

THE IMPACT OF FAMILY COMPLEXITY ON THE TREATMENT  
OUTCOME OF BEHAVIOURAL SLEEP INTERVENTION IN CHILDREN  
WITH AUTISTIC SPECTRUM DISORDERS

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By Yvonne Wai-Yi CHOW

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## **Abstract**

Sleep problems affect not only the wellbeing of children with autism spectrum disorders (ASD) but also the functioning of the entire family unit, and bi-directional influences between child sleep and family functioning are possible. Complex familial factors are seldom considered as predictors of outcome in studies on ASD. The present study aimed at investigating the effect of a Functional Behavioural Assessment (FBA) informed parent-implemented behavioural sleep treatment on ameliorating sleep problems of children with ASD. The present analysis also investigated the impact of components of family complexity on sleep problem severity in these children, in particular how well these components could predict treatment outcomes. To achieve this, the present analysis explored the components that lay underneath the 11 parent/family variables used to operationally define family complexity.

Data from thirty-one children with ASD, aged between 3 and 14.5, and their parents, were analysed. These 31 children completed the treatment phase of a larger study (54 participants) which investigated the effectiveness of a 4-to-8- week home-based behavioural sleep intervention in ameliorating sleep problems in children with ASD. Exploratory factor analysis was used to explore the components that reside underneath the complex family dynamics. Multiple regression analysis was utilised to investigate whether components of family complexity could predict treatment outcomes.

The results of pre- and post-comparisons indicated the efficacy of behavioural sleep treatment in reducing sleep problem severity in the children at post-treatment, short-term follow-up and long-term follow up. Secondary outcomes included improved paternal depression, paternal anxiety, maternal anxiety and maternal sleep

quality. Factor analysis indicated there were three components of family complexity. The three components were paternal wellbeing (paternal mood states and paternal sleep quality), maternal wellbeing (maternal mood states and maternal sleep quality), and parental relationship quality (paternal marital satisfaction and maternal marital satisfaction). Parental relationship quality before treatment was found to be the only significant predictor of treatment outcomes,  $F(3,19) = 3.86, p = .026$ .

The evidence of the present study provided stronger support for a bi-directional relationship between child sleep and parental wellbeing. Future research efforts could consider examining the joint effect of child factors and family factors in predicting the treatment outcomes in children with ASD.

## **Chapter 1**

### **Introduction**

Sleep problems (e.g., difficulties initiating and maintaining sleep) are common in children with autism spectrum disorders (ASD), particularly when compared to typically developing (TD) children and those with other types of developmental disabilities (Polimeni, Richdale, & Francis, 2005). Data collected from parent-report indicates that the prevalence of sleep problems is between 54% - 89% for children with ASD compared to 25% - 40% for TD children (Allik, Larsson, & Smedje, 2008; Couturier et al., 2005; Didden & Sigafos, 2001; Malow, Marzec, McGrew, Wang, Henderson & Stone, 2006; Polimeni et al., 2005; Richdale, Francis, Gavidia-Payne & Cotton, 2000), and 24-68% for children with other developmental disabilities (Didden & Sigafos, 2001; Polimeni et al., 2005; G. Stores & R. Stores, 2013; Wiggs & Stores, 2004 ).

Children with ASD might be more vulnerable to the influence of disrupted sleep than TD children (Souders et al., 2009). Previous studies have suggested that sleep problems may aggravate the core symptoms of ASD, including decreased emotional and psychosocial wellbeing (Kuhlthau et al., 2010), reduced social communication skills (e.g., impaired attention; Malow & McGrew, 2008), reduced nonverbal intelligence, adaptive behaviour and motor skills (Taylor, Schreck, & Mulick, 2012), increased rates of restricted and repetitive behaviours (e.g., preservative interests; De Vincent, Gadow, Delosh, & Geller, 2007; Richdale et al., 2000), increased daytime behaviour problems, such as social skills deficits, hyperactivity, repetitive behaviours, heightened aggression, and self-injury (Goldman et al., 2011; Fadini et al., 2015; Maski & Kothare, 2013).

Sleep problems not only affect children with ASD but also the functioning of the entire family unit (Lyons, Leon, Phelps, & Dunleavy, 2010). The quality of parental wellbeing (Doo & Wing, 2006), marital relationship (Wiggs & Stores, 2004), and sleep of other family members (Chou et al., 2012; Melzer, 2008) could be affected. The disrupted sleep of children with ASD is likely to reduce the wellbeing, interpersonal relationship and sleep quality of the parents (Richdale & Schreck, 2009).

Without effective treatment, sleep problems might become chronic and persist through adolescence and into adulthood (Sivertsen, Posserud, Gillberg, Lundervold & Hysing, 2012). Identifying treatment non-responders is, therefore, an important concern in developing new treatments that help those children who do not benefit from current therapy protocols. Although it is crucial to develop treatment with enhanced efficacy, the factors that alter the efficacy of a treatment also need to be identified. Questions, such as in which child an intervention is effective or in what circumstances an intervention is insufficient to help a family, have received much concerns in recent years in child psychotherapy studies (Brestan & Eyberg, 1998; Owens et al., 2003). Therefore, it is important to understand what family factors could influence treatment outcomes in children with ASD. Identification of reliable predictors of success and failure of treatment for families may increase the clinicians' awareness of what specific familial difficulties in treatment planning need to be addressed so as to minimise the cases of non-responders (Webster-Stratton, 1985).

It is also critical that we not only identify effective treatments but also enhance our understanding of the social validity of sleep treatment, parental experiences with the assessment and treatment process that may be related to the therapeutic responses of the child (Elliot, 1988). Yet within child behavioural sleep treatment research,

rarely have studies examined parent or family factors that influence treatment outcomes (Kazdin, 1995).

Very few studies have examined the relationship between child characteristics (e.g. autism severity and social skills) and treatment outcomes in children with challenging behaviour (Kazdin & Whitley, 2006; Pellecchia et al., 2015). There are also limited studies that have identified family factors as predictors on therapeutic outcome in children with conduct disorder (Webster-Stratton, 1985; Webster-Stratton & Hammond, 1990). Not much is known, however, about the impact of familial characteristics (e.g., parental mood states or interparental relationships) on treatment outcomes. The present study investigated the impact of family complexity (i.e., parental mood states, parental sleep quality, parental marital relationship and family adversity) on sleep problem severity and treatment outcomes in children with ASD.

### **Importance of Considering Sleep within a Family Context**

To address sleep within a family context is one of the critical dimensions of sleep research which has been commonly absent from discussions in current sleep-related studies (Dahl & El-Sheikh, 2007). The Family System approach emphasises the interactions within a family, such as the communications among the mother-child, marital and sibling subsystems (Bowen, 1985; Kerr & Bowen, 1988). Urie Bronfenbrenner also used the bio-ecological system model to explain how a child's inherent qualities and his environment interact with one another to influence how he/she will grow and develop (Bronfenbrenner, 1999). Bronfenbrenner highlighted the importance of understanding a child in the context of multiple environments, also known as ecological systems (Bronfenbrenner & Crouter, 1983). Bronfenbrenner (1977; 1979; 1986) used four ecological subsystems (i.e., four layers) to explain how external factors have proximal or distal influences on the development of a child. The

microsystem is the innermost level (of the ecological systems) that includes the child's immediate family, peers, school, and neighbourhood. Proximal processes are likely to occur in this subsystem. Bronfenbrenner (1999) regarded the processes in microsystem are bi-directional because the family can affect the child's behaviour whereas the child's temperament, deficits or behaviour can influence the responses of the others. The mesosystem is the second immediate layer that emphasises the connections between two or more microsystems, for example, the parent-teachers relationship (Bronfenbrenner, 1999). The exosystem is the third layer with which a child does not directly interact. However, it could impact his/her development through distal processes. The examples of exosystem are family social class, work context of the parents, and social services that a family can access (Bronfenbrenner, 1999). The outermost layer is the macrosystem which includes cultural beliefs and values that might have distal or indirect influences on a child (Bronfenbrenner, 1999).

Family-focused ASD research is the key gateway to increasing our understanding of the impact of family factors on children with ASD as well as to informing applicable clinical support services for the affected families (Cridland, Jones, Magee & Caputi, 2014). There are several reasons to consider the mutual influence of children's sleep and family factors. Firstly, the sleep problems of a child often affect the other family members. For instance, the night awakening of a child can have a significant impact on parental sleep, marital relationships and daytime functioning of the parents (Meijer & van den Witenboer, 2007). Secondly, family contexts such as family structure bedtime rituals, cultural practices, parental attitudes and beliefs about sleep and home arrangement also have marked influence on the sleep of a child (Milan, Snow, & Belay, 2007; Owens, 2004). Thirdly, the treatment outcomes of sleep intervention in children are affected by the parental expectations of



treatment effectiveness and the parental perceptions regarding ‘normal’ sleep in children (Owens, 2004; Robinson & Richdale, 2004). Thereby, the sleep problems among children should not be discussed in isolation from a family context. Studies focusing on family context are essential for increasing our understanding of how to best support families having children with ASD.

### **Autism Spectrum Disorders**

**Definition.** Autism spectrum disorders are a cluster of neurodevelopmental disorders characterised by impairments in social-emotional reciprocity, verbal/nonverbal communication and flexible imaginative functions (e.g., restricted interests, stereotyped and repetitive behaviours) (American Psychological Association, APA, 2013). In the DSM-5, ASD includes autistic disorder and social (pragmatic) communication disorder (APA, 2013). In children with ASD, the symptoms typically present in the first two years of life. The severity of ASD can range from mild to profound depending on the developmental level, cognitive ability (e.g., mental retardation) and associated behavioural symptoms (e.g., hyperactivity, aggressiveness and temper tantrums) that a child manifests.

**Prevalence of ASD.** National estimates reported that, in the US, 1 in 68 (1.5%) children are affected by ASD (Autism and Developmental Disabilities Monitoring Network Surveillance Year, 2010). Although New Zealand (NZ) based data is limited, in 2008, it was estimated that approximately 40,000 individuals in NZ had a diagnosis of ASD (Ministry of Health, 2008). This prevalence appears to be rising. In 2015 a national survey conducted in NZ revealed that about 1.2% of children (approximately 11,000 children) between 2 to 14 years of age had ASD (Ministry of Health, 2015). The prevalence rate increased to 1.4% in 2016 (Ministry of Health, 2016) and climbed up to 2.2% (approximately 18,000 children) in 2017

(Ministry of Health, 2017). ASD is 4.78 times more common in boys than in girls (Ministry of Health, 2017). Elevated exposure to drugs and other toxins during prenatal and perinatal stages, remarkable advances in our understanding and recognition of ASD (Chaste & Leboyer, 2012), and diagnostic substitution are all factors that may contribute toward the rising prevalence rates of ASD in NZ and around the globe.

**Aetiology of ASD.** The cause of ASD is still unknown; however, some researchers suggest that ASD is the product of an interaction between genetic and environmental factors (Grabrucker, 2013).

**Genetic factors.** Studies into twins reported an 88% median concordance rate of monozygotic twins, suggesting a high heritability of ASD (Ronald & Hoekstra, 2011). Higher rates of ASD among siblings of children with this diagnosis are also thought to be evidence of a genetic component to this disorder. Recent research suggests that the reported risk of ASD recurrence in families is at least 4–7% (Blenner, Reddy & Augustyn, 2011). Although there is no consensus in the field about which genes are involved in ASD, researchers suggest that the duplication of chromosome 15q11-13 is related to the characteristics of ASD (Bundey, Hardy, Vickers, Kilpatrick, & Corbett, 1994; Cook et al., 1998). Subsequent studies have also provided evidence that the cytogenetic abnormalities in maternal duplications of the imprinted domain on chromosome 15q11–13 might contribute to ASD in children (Grafodatskaya, Chung, Szatmari & Weksberg, 2010; Schanen, 2006). In another study, Gregg et al. (2008) have identified 11 genes (e.g. PAM, SPON2, & IL2RB) that are differentially expressed in the ASD groups compared to those in the TD control cohort. These findings, though tentative, suggest that there may be a genetic predisposition to ASD.

***Environmental factors.*** So far, there is no conclusive empirical evidence, regarding the significance of environmental factors contributing to ASD, but studies suggest some common features that often link to the affected children. In a review article, Karimi and colleagues (2017) summarised a list of prenatal risk factors that would increase a child's susceptibility to ASD. The risk factors included the use of drugs (e.g., antidepressant), exposure to toxic substances (e.g., pesticides), older parental age and low familial SES (Karimi, Kamali, Mousavi & Karahmadi, 2017). Some studies have demonstrated that prenatal exposure to air pollution (e.g., heavy metals and particulate) also increases the risk of conceiving children with ASD (Volk, Hertz-Picciotto, Delwiche, Lurmann & McConnell, 2011; Volk, Lurmann, Penfold, Hertz-Picciotto, & McConnell, 2013). Nevertheless, it is important to note that some researchers were unable to find a significant correlation between prenatal exposure to environmental toxicity and the presence of ASD in newborns (Kalkbrenner et al., 2010); thus further research focusing on familial or environmental factors is required.

***Epigenetic factors.*** Despite the evidence of environmental and genetic contributors, the majority of individuals with ASD are likely to have idiopathic autism with no identifiable genetic deficit or environmental similarities (Karimi et al., 2017). Nevertheless, it is commonly believed that ASD may be the result of epigenetic factors. Bronfenbrenner has also suggested that effective psychological functioning and actualisation of genetic potentials rely on the synergistic effects of genetics-environment interaction (Bronfenbrenner & Ceci, 1994). Atypical epigenetic mechanisms and gene expression (e.g., chromosomal copy number variants; CNVs) are often found in individuals with neurodevelopmental disorders (Gropman & Batshaw, 2010). The CNVs in chromosomal region 16p11.2 were found related to increased time awake and reduced NREM sleep associated with ASD (Angelakos et

al., 2017). Moreover, environmental insults such as maternal viral infection and maternal bacterial infection in the prenatal period may interact with the ASD-associated susceptibility genes during the processes of synaptogenesis, which leads to a possible alteration in gene expression resulting in an increased risk of ASD (Atladóttir et al., 2010; Blenner et al., 2011; Nardone et al., 2014). Recent genetic studies have reported that children with ASD who had CNVs and exposure to prenatal maternal infection demonstrated more social communicative difficulties and restricted/repetitive behaviours compared with those children with ASD who were exposed to maternal infection but did not have CNVs (Fakhoury, 2015; Mazina et al., 2015). These findings suggest that ASD might be a product of the interacting effect of genetic and environmental factors (Schaevitz & Berger-Sweeney, 2012).

### **Importance of Sleep to Humans**

**Definition of sleep.** Sleep is defined as a complex behavioural and physiological state of perceptual disengagement from and unresponsiveness to the environment (Carskadon & Dement, 2011; Mindell & Owens, 2015). It can be characterised by minimal motor activity, reduced interaction with and responsivity to the environment, specific gestures (e.g., lying down with closed eyes) and easy reversibility (Carskadon & Dement, 2011; Mindell & Owens, 2015).

**Functions of sleep.** Sleep is essential for maintaining daily physical, emotional and cognitive functioning (Plywaczewski et al., 2003). Some theories have tried to interpret the function of sleep as a restful time for physical recovery that enables the body to grow and repair, but much remains unknown. Schmidt (2014) has proposed an energy allocation model of sleep, which is a unifying theory considering sleep as an energy conservation strategy based on the assumption that the body needs to optimally distribute limited energy resources to vital biological functions such as

tissue restoration, growth, maintenance and reproduction. According to this theory, sleep re-allocates energy utilisation away from the high demands of wakefulness (e.g., skeletal muscle tone) into other essential biological operations (e.g., central nervous system functions) when an individual is not engaged in waking-related activities. Haack and Mullington (2005) investigated whether an individual's sleep, restricted to 50% of the habitual time over twelve days, had an impact on the subjective ratings of physical symptoms. The data revealed that there were increases of generalised body tiredness and fatigue, back pain and stomach discomfort across days of sleep deprivation.

Besides energy conservation, sleep also helps emotional regulation. Sleep deprivation has been found related to anxiety, depression and affective psychopathology (Baglioni, Spiegelhalder, Lombardo & Riemann, 2010; Franzen, Buysse, Dahl, Thompson & Siegle, 2009). The amygdala has a significant role in processing emotional information, particularly aversive feelings (Sotres-Bayon, Bush, & LeDoux, 2004). The medial prefrontal cortex (MPFC) is proposed to exert top-down inhibitory control of amygdala function contributing to contextually appropriate emotional reactions (Davidson, 2002). As little as twenty-hour continuous wakefulness is correlated with significant reductions in blood flow and glucose metabolism within the prefrontal cortex (Thomas et al., 2000). Yoo et al. (2007) reported that, in comparison to the sleep-deprived adults, there was significantly stronger connectivity between the amygdala and the MPFC in the sleep-control group. The result suggests that sleep deprivation might predict the failure of top-down, prefrontal control of amygdala function that subsequently leads to the elevated risk of emotional dysregulation.

It has been reported that sleep might play a role in memory consolidation and learning (Siegel, 2001). Regarding memory and learning consolidation, it is necessary for the information learned during the day to be transferred between the cerebral cortex and the hippocampus during sleep (Wilson & McNaughton, 1994). Slow-wave sleep might facilitate the flow of information from the hippocampus to the neocortex (Buzsáki, 1996; Chrobak & Buzsáki, 1994). The rapid eye movement (REM) sleep-memory consolidation hypothesis has received support from data indicating the REM-dependent development of binocular cells evident in the visual cortex, a brain region which is crucial for learning to occur (Frank, Issa & Stryker, 2001).

### **Architecture of Sleep**

Researchers of sleep studies have examined the sleep architecture of individuals with ASD (Elia et al., 2000; Buckley et al. 2010). The findings suggest that individual with ASD possibly has sleep architecture different from typical sleep architecture (Buckley et al., 2010; Elia et al., 2000; Lopez et al. 2008). Some evidence has revealed that the atypical sleep architecture of children with ASD might be related to familial factors (Engelhardt, Mazurek, & Sohl, 2013; Mazurek, Engelhardt, Hilgard, & Sohl, 2016).

**Typical sleep architecture.** The framework of sleep is composed of three sleep states: awake, non-rapid eye movement (NREM) sleep, and REM sleep. These stages are characterised by distinct polysomnographic features of electroencephalographic (EEG) patterns, eye movements and muscle tone. The non-rapid eye movement sleep is identified by large amplitude and low-frequency EEG oscillations (Carskadon & Dement, 2011; Rechtschaffen & Kates, 1968). NREM consists of four stages in traditional classification (Rechtschaffen & Kates, 1968), and has been modified to 3 stages more recently (i.e., NREM 1, 2 & 3) that are roughly

parallel to a depth continuum of sleep, with arousal thresholds generally lowest in Stage 1 and highest in Stage 3/4 sleep (Carskadon & Dement, 2011).

The REM sleep, also known as paradoxical sleep, active sleep, dream sleep and slow wave sleep, is characterised by low-amplitude, fast rhythms on EEG recordings, rapid eye movements behind closed eyelids, and temporary muscle paralysis (Carskadon & Dement, 2011). Over the night, while NREM 2 keeps expanding from the second NREM-REM cycle onwards, NREM 3 and 4 diminish during the second NREM-REM cycle (Mindell & Owens, 2015). The duration of REM sleep episodes typically increases across the night.

Starting from the age of two, a child's sleep onset begins to occur through NREM stages rather than REM sleep. NREM 3 and 4 sleep decrease across adolescence by 40% from preadolescent years and continue to deplete slowly into adulthood. REM sleep occupies 20-25% of sleep, occurring in four to six discrete episodes throughout a night, across childhood, adolescence, adulthood, and into old age. NREM sleep, therefore, contributes more than two-thirds of one's sleep (Carskadon, & Dement, 2011).

**Atypical sleep architecture in ASD individuals.** Elia et al. (2000) recorded the atypical architecture of sleep in children with ASD. This study revealed that the first REM sleep latency appeared to be shorter in ASD samples than in TD controls, although this difference did not reach statistical significance. The time of NREM 1 sleep was found shorter in the ASD group when compared with children with developmental disorders (DD). The ASD group also demonstrated more muscle twitches during sleep (Elia et al., 2000). Buckley et al. (2010) compared the polysomnographic recordings between children with ASD, TD children and children with DD. The ASD group manifested a significantly shorter sleep time in total with

more slow wave sleep and much lesser REM percentage than the other two groups. However, in contrast to the findings of Elia et al. (2000), the ASD samples in the research of Buckley et al. (2010) had a greater percentage in NREM 1 sleep. Despite this inconsistent result, the REM sleep deprivation in the ASD samples of both studies may imply there might be a different sleep architecture associated with sleep difficulties in ASD (Lopez et al. 2008).

The atypical sleep profiles of ASD children could be related to familial factors such as bedtime routines and excited bedtime activities (Stores & Wiggs, 1998). Evening media use and playing exciting computer games were associated with decreased total sleep time, increased sleep latency and reduced REM sleep in TD children and adults (Garrison, Liekweg, & Christakis, 2011; Higuchi, Motohashi, Liu, & Maeda, 2005; Oka, Suzuki, & Inoue, 2008). Compared with TD children, children with ASD spent more hours on bedtime media use, which was found correlated with greater sleep onset latency (Engelhardt, Mazurek, & Sohl, 2013; Mazurek, Engelhardt, Hilgard, & Sohl, 2016). It has been reported that increased access to screen-based media and reduced parental restriction over media use were correlated with increased oppositional behaviour and reduced total sleep time in boys with ASD (Engelhardt & Mazurek, 2014; Engelhardt, Mazurek, & Sohl, 2013). These findings suggest that family factors might alter a child's sleep architecture.

### **The Importance of Sleep in Child Development**

Sleep serves multiple functions in child development, including physical growth, neuronal development, memory processing, and cognitive maturation (Stores & Wiggs, 1998). In children with ASD, sleep deprivation has demonstrated not only an exacerbation to the severity of ASD symptoms (e.g., repetitive behaviours, social and communication deficits; Park et al., 2012; Tudor et al., 2012), but also negative



effects on cognitive functioning, emotional regulation and daytime behaviours (e.g., self-injury, tantrums, and aggression; Goldman et al., 2011; Henderson, France, Owens, & Blampied, 2010).

**Cognitive functioning.** Sleep is essential for learning, memory processing and academic performance for children and adolescents. Research revealed that inadequate sleep, increased sleep disruption, late bedtimes and early awakenings could impair a child's learning ability, academic performance and intellectual functioning (Dewald et al., 2010). For example, fluid reasoning, a kind of problem-solving intelligence, is affected when a child does not get enough REM sleep (Volk & Huber, 2015). Experimental sleep manipulation studies in children are sparse. Nonetheless, some evidence suggests that sleep deprivation among TD children can negatively affect their verbal creativity, abstract reasoning, and academic performance (Fallone, Acebo, Seifer, & Carskadon, 2005; Randazzo, Muehlbach, Schweitzer, & Waish, 1998).

Findings from studies of children with ASD have suggested that sleep deprivation and sleep fragmentation are correlated with impaired intellectual functioning, low IQ, nonverbal intelligence deficits, verbal skills difficulties, and poor academic performance (Karpinski, Scullin & Montgomery-Downs, 2008, Paavonen, Nieminen-von Wendt, Vanhala, Aronen, & von Wendt, 2003, Taylor et al. 2012).

**Socioemotional regulation and daytime behaviour.** There is increasing empirical evidence substantiating the role of sleep in socioemotional regulation. Literature reviews have reported a link between sleep deprivation and impaired emotional regulation in TD children (Beebe, 2011; Gregory & Sadeh, 2012) and children with ASD (Kotagal & Broomall, 2012). Beebe (2011) reviewed thirteen relevant studies and summarised the findings that childhood sleep difficulties play a

critical role in the development of mood disturbance over time, even after controlling the baseline mood disturbance and other confounding variables. Moreover, this situation has been reported spanning different time frames, namely, preschool to mid-childhood (Jasper et al., 2010, Gregory, Eley, O'Connor, & Plomin, 2004), preschool to mid-adolescence (Gregory & O'Connor, 2002; Wong, Brower, & Zucker, 2009), mid-childhood to late childhood (El-Sheikh, Kelly, Buckhalt & Benjamin, 2010; Gregory, Rijdsdijk, Lau, Dahl, & Eley, 2009), mid-childhood to young adulthood (Gregory, Van der Ende, Willis, & Verhulst, 2008), and adolescence to young adulthood (Roane & Taylor, 2008). Although the longitudinal associations between sleep problems and externalising behaviours have been reported, the correlation tends to be weaker or less consistent (El-Sheikh et al., 2010, Gregory et al., 2004, Gregory et al., 2008, Jasper et al., 2010) than those observed in internalising disorders. For example, in a study analysing the behavioural difference between a group of TD adolescents' healthy sleep days (i.e., 10 sleep hours/night) and restricted sleep days (i.e., 6.5 sleep hours/night), it was discovered that they experienced elevated anxiety, hostility, confusion, fatigue, and reduced energy ( $p = .001$  to  $.01$ ) after undergoing the days in which sleep was restricted (El-Sheikh et al., 2010). Parents and adolescents also reported experiencing greater oppositionality/irritability and poorer emotional regulation during the restricted sleep days. By contrast, differences in hyperactivity/impulsivity were not significant (El-Sheikh et al., 2010).

In children with ASD, sleep problems were found positively correlated with hyperactivity, tantrums, aggressive behaviour, stereotypic behaviour, emotional problems, inattentive and hyperactivity, irritability and challenging behaviours (DeVincent, Gadow, Delosh, & Geller, 2007; Goldman et al., 2009; Henderson,

Barry, Bader, & Jordan, 2011; Park et al., 2012; Malow et al., 2006; Mayes & Calhoun, 2009).

**Adaptive functioning.** Taylor et al., (2012) reported that, compared to the well-rested children, children with ASD who were sleep-deprived (i.e., sleeping fewer hours per night) displayed more difficulties in adaptive functioning, daily living skills, social ability, and motor ability. Children who had more night wakings (e.g., screaming) and higher sensitivity to environmental stimulus at bedtime (e.g., sound) were more prone to incompetence in daily living skills (Taylor et al., 2012). Among children with ASD, those with sleep disturbance were found more likely to have withdrawal, social and communication difficulties compared with those without sleep disturbance (Cohen, Conduit, Lockley, Rajaratnam, & Cornish, 2014; DeVincent et al., 2007; Goldman et al., 2009).

**Physical functioning.** The processes of growth and development are believed to be related to sleep. Evidence has emerged that deep sleep triggers the release of growth-promoting hormones, which boosts muscle mass and promotes metabolic milieu in children (Miller, Lumeng, & LeBourgeois, 2015; Snell, Adam, & Duncan, 2007). Short sleep duration has been repeatedly identified as a risk factor for childhood obesity in cross-sectional (Cappuccio et al., 2008) and longitudinal studies (Chen, Beydoun, & Wang, 2008; Collings et al., 2016; Lytle et al., 2013; Martinez et al., 2014). For example, significant associations between sleep, body mass index and overweight condition of children were discovered in a study with longitudinal data from a sample of 2,281 TD children (3 to 12 years old; Snell et al., 2007).

These findings have suggested that poor sleep quality or inadequate sleep quantity can lead to immediate and long-term cognitive, behavioural, functional and physical consequences in child development (Beebe, 2011).

## **Prevalence of Sleep Problems in Children with ASD**

Sleep problems are among the most common worries for parents of children with ASD. While estimates vary, sleep problems are reported occurring in 54-89% of children with ASD (Doo & Wing, 2006; Hoffman et al., 2006; Liu, Hubbard, Fabes & Adam 2006; Richdale, 1999; Wiggs & Stores, 2004). By contrast, the same problems are reported happening only 25-40% in TD children (Krakowiak et al., 2008; Owens & Moore 2017; Meltzer & Mindell, 2008; Polimeni et al., 2005; Reynolds & Makow, 2011), 20-50% in children with ADHD (Corkum, Tannock, & Moldofsky, 1998; Meltzer & Mindell, 2008 ) and 24-68% in children with other developmental or intellectual disabilities ( Didden & Sigafoos, 2001; Polimeni et al., 2005; G. Stores & R. Stores, 2013; Wiggs & Stores, 2004 ).

In one study, Liu, Hubbard, Fabes and Adam (2006) interviewed the parents of 167 children with ASD (mean age 8.8 years). It has been found that about 54% of these children demonstrated bedtime resistance, 56% suffered from insomnia, 53% experienced parasomnias, 25% showed sleep-disordered breathing, 45% had morning rise problems, and 31% reported daytime sleepiness. Krakowiak et al. (2008) compared the sleep patterns of 3 groups of children: TD children, children with ASD, and children with developmental delay. More than 50% of children with ASD showed at least one frequent sleep difficulty, followed by 46% of the developmental delay group, and 32% of TD children ( $p < .001$ ). The children with ASD experienced significantly more frequent night wakings ( $p < .001$ ) and difficulties with sleep onset ( $p < .001$ ) than the TD group (Krakowiak et al., 2008). Similar findings have been reported in some additional studies (Gail, Sears & Allard, 2004; Polimeni et al., 2005; Wiggs & Stores, 2004).

Although some studies have demonstrated that children with ASD are more

vulnerable to sleep difficulties, the difference in sleep patterns between children with ASD and TD children have not been consistent. Schreck and Mulick (2000) examined the sleep patterns of 169 children with ASD (ages 5 to 12) and found that their average nighttime sleep duration was about 9.0 hours, which was not significantly different from the total sleep time of TD children. Interestingly, although the sleep quantity of the children with ASD was found to be somewhat reasonable, their parents, compared with the parents of TD children, were more likely to perceive that their children had disrupted sleep. Similar findings were also reported by Hering, Epstein, Elory, Iancu & Zelnik (1999). In this study, parents reported that their child with ASD experienced early morning wakings and frequent nighttime awakening. However, the actigraphic recordings revealed that, except for an earlier morning arousal time ( $p = .045$ ), sleep patterns of children with ASD were similar to that of TD children. Moreover, TD children and children with ASD had a similar quantity of sleep. It is possible, therefore that the parents of children with ASD overestimate and over-react to the sleep disturbance of their children. Children who do not sleep well might demonstrate more daytime challenging behaviours than children without sleep disturbance (Wiggs & Stores, 1996). Poor sleep is associated with exacerbated disruptive and difficult behaviours in children with ASD (Allik, Larsson & Smedje, 2006; Richdale & Schreck, 2009). Children with ASD have been reported to engage in higher levels of challenging behaviour than TD children ( $p < .001$ ; Matson, Wilkins & Macken, 2008). Parents of children with ASD often experienced elevated levels of parental stress after handling the challenging behaviours of their children, which might make the stressed parents over-sensitive to the sleep problems of their children (Baker et al., 2003; Davis & Carter, 2008).

Hastings (2003) revealed that, compared with fathers, mothers were more

affected by children's self-regulation problems such as sleeping and emotion regulation. Research revealed that mothers are likely to be more involved in daily care of their children, especially those with children with ASD; thereby sleep problems of children tend to be more impactful on the mothers. Davis and Cater (2008) studied the parental mental wellbeing in 54 families having children with ASD. They reported that the parenting stress and depression levels of the mothers were generally higher than the fathers'. Many past studies of ASD associated problems have been criticised for using the mothers' reports about their children's daily behaviours primarily. Analysis indicated that the predominance of using the data about children's behaviour from the mothers' questionnaire, particularly the sleep behaviour, may incur immoderate negative perceptions of a child's sleep problems (Davis & Cater, 2008). This might make the sleep problems of children with ASD appeared to be more severe.

### **Types of Sleep Disorders**

According to the third edition of the International Classification of Sleep Disorders (ICSD-3), there are seven major categories of sleep disorders; namely, insomnia disorders, sleep-related breathing disorders, central disorders of hypersomnolence, circadian rhythm sleep-wake disorders, sleep-related movement disorders, parasomnias, and other sleep disorders (American Academy of Sleep Medicine, 2014). Among them, insomnia, parasomnia, and circadian rhythm sleep-wake disorders are the three most reported sleep disorders in children with ASD (Cohen et al., 2014; Fricke-Oerkermann et al., 2007), and they are among the top parental concerns reported to clinicians (Mindell, Moline, Zendell, Brown, & Fry, 1994).

**Insomnia.** Insomnia refers to persistent difficulty with sleep initiation, maintenance, duration, consolidation, or quality. Subtypes of insomnia include chronic insomnia, short-term insomnia and non-specified insomnia (Sateia, 2014). The behavioural insomnia of childhood is included in the chronic insomnia disorder diagnosis. Research reported that behavioural insomnia was found evident in 66% of children with ASD compared to 45.9% in controls (Souders et al., 2009). There are two main types of behavioural insomnia: sleep-onset association and limit-setting (Sateia, 2014). Sleep-onset association results from the negative associations of environmental factors (e.g., the presence of parents) with sleep. These children commonly require a parent or soothing item (e.g., toy) to help them sleep. Limit-setting type refers to the children's challenge to their parents during bedtime and refusal to go to sleep. This can often be seen in either resisting going to bed on time, or making many requests (e.g., snacks) so as to delay sleeping (Sateia, 2014).

**Parasomnias.** Parasomnias are classified into three subtypes: NREM related (e.g., sleepwalking and sleep terrors), REM-related (e.g. nightmare disorder), and non-specified parasomnias (Sateia, 2014). Children who suffer from parasomnias often complain of having aversive physical or psychological experiences such as nightmares, wake screaming, complex movements and unpleasant dreams that occur while sleeping or during the arousal from sleep (Cohen et al., 2014). It has been found that 54% of children with ASD had multiple and early night arousals due to parasomnias compared to 5% to 35% in TD children (Hering et al., 1999; Liu, Liu, Owens, & Kaplan, 2005).

**Circadian rhythm sleep-wake disorders.** Circadian rhythm sleep-wake disorders refer to the alterations of the circadian time-keeping system, the failure in 24-hour entrainment or misalignment of the internal body clock and the

environmental stimulus (e.g., light; Cohen et al., 2014). The individuals having circadian rhythm sleep-wake disorders are unable to fall asleep at a normal bedtime. Subtypes include delayed sleep-wake phase, advanced sleep-wake phase and irregular sleep-wake rhythm (Sateia, 2014). Approximately 10% of children with ASD were found having sleep-wake rhythm problems that varied with season due to day-night changes (Giannotti et al., 2008).

### **Common Sleep Disturbance in Children with ASD**

Some children with ASD might not be diagnosed with having the above-listed sleep disorders but still, have problems with sleep. These children might display some symptoms of sleep disorders, but the severity does not reach a clinical diagnostic level. It is possibly because of the atypical sleep architecture (e.g., shorter total sleep time, more muscle movements), it is not surprising that parents of children with ASD complain about their children having sleep problems such as difficulty falling asleep, frequent night wakings, short sleep duration, restless sleep, parental presence request, and co-sleeping (Humphreys et al., 2014; Richdale & Schreck, 2009; Wiggs & Stores, 2004).

**Settling difficulty and sleep onset latency.** The settling difficulties of children with ASD may include bedtime resistance, bedtime routine non-compliance (Krakowiak et al., 2008, Malow et al., 2006) as well as frequent curtain calls (i.e., bids for parental attention; Allik et al., 2006; Bruni et al., 2007; Patzold et al., 1998). Bedtime refusal may lead to delayed sleep onset. Sleep onset latency (SOL) is the length of time that an individual needs for the transition from full wakefulness to falling into a light sleep, usually the first episode of NREM sleep (Krakowiak et al., 2008). If the duration of a sleep onset latency is more than 20 minutes for a child to fall asleep after going to bed, it is regarded as a delayed sleep onset (Owens, Spirito,



McGuinn, & Nobile, 2000). Studies have reported that children with ASD could take more than 60 minutes to fall asleep (Krakowiak et al., 2008; Malow et al., 2006).

**Night waking.** Night waking refers to arousal occurring during a night in which a child does not self-soothe to fall asleep again (Henderson et al., 2010). Both sleep quality and sleep quantity are affected by frequent and prolonged night wakings (i.e., fragmented sleep; Ohayon et al., 2017). Not all children could sleep continuously for 10 to 12 hours per night. A review article indicated that, for those aged 3 to 17, a night-time awakening after sleep onset with a duration between 21 and 50 minutes might reduce the sleep quality. An awakening of more than 51 minutes for pre-schoolers and teens and a night-time arousal of longer than 41 minutes for school-aged children denote poor sleep quality (Ohayon et al., 2017).

It has been revealed that children with ASD often wake up during the night or early in the morning (Allik et al., 2008, Souders et al., 2009, Wiggs & Stores, 2004, Williams, Sears, & Allard, 2004). Night wakings are regarded as problematic behaviours by parents, particularly when the arousals involve signalling (i.e., the child cries, refuses to sleep, or gets out of bed), and are recurring and prolonged (Cortesi, Giannotti, Ivanerko, & Johnson, 2010). Studies utilising actigraphy and sleep diary have revealed the duration of night waking in children with ASD can last up to two to three hours. During this time children were commonly observed laugh, self-talk, cry, or get up and play with objects (Cortesi et al., 2010, Giannotti et al., 2008).

**Early rise times.** Many parents considered the behaviour of early awakenings in their children with ASD problematic (Richdale & Prior, 1995). The early awakening is defined as an inappropriate time of arousal that a child wakes up earlier than the time that a family deemed as appropriate for the child to wake up for day commencement (Richdale & Prior, 1995). Early awakening has been reported being

more common among children with ASD than their TD peers (Malow et al., 2006). Early rising can negatively affect parental sleep and can result in inadequate sleep duration for both children and their parents (Meltzer, 2008; Richdale & Prior, 1995).

**Reduced total sleep time.** Goodlin-Jones, Tang & Anders (2008) defined an individual's total sleep time as the duration of the actual sleeping time in a sleep episode; which is equal to the duration of night waking subtracted from the duration of total sleep episode. Sleep duration is likely to be reduced as a consequence of delayed sleep onset, multiple night wakings or early arousal. Goodlin-Jones et al. (2008) revealed that the total sleep time (10hrs 36mins) of children with ASD (2-5.5 years old) was found less than the TD children (11hrs 14mins) and the children with DD but without ASD (11hrs 6mins).

**Parental presence during sleep onset.** Parental presence is often defined as the parental involvement in which a parent stays in the child's room providing reassurance to help the child fall asleep (Karitane and Tresillian, 2015). Parental involvement includes a parent being in physical or visible proximity of the child and engaging in caring behaviours such as nursing the child to sleep or laying with the child (Meltzer & Mindell, 2006). Children with ASD are often dependent upon the presence of a parent in order to initiate and maintain sleep (Wiggs & Stores, 2004). The presence of a parent during bedtime can be problematic because it might strengthen the association between sleep and physical contact with the caregiver. The child would find it difficult to sleep through a night if the parent is not nearby (Meltzer & Mindell, 2006). Studies have reported that infants who have prolonged physical contact (e.g., nursing or rocking the child to sleep or co-sleeping) with parents when settling to sleep demonstrate a higher frequency and duration of night waking than infants whose parents restrain from such physical contact behaviours

during sleep onset (Burnham, Goodlin-Jones, Gaylor, & Anders, 2002; Mao, Burnham, Goodlin-Jones, Gaylor, & Anders, 2004). Wiggs & Stores (2004) studied the sleep problems of a group of children with ASD (5 to 14 years old) and reported that more than 25% of them frequently requested for parental presence (at least 3 times a week) at sleep onset or following night wakings.

**Co-sleeping.** McKenna and Volpe (2007) defined co-sleeping as the presence of at least one caregiver who sleeps closely to the child enabling the exchange of at least two sensory stimuli (e.g., auditory, olfactory and tactile). Despite the opposition from western medical authorities or policy officials, co-sleeping is more acceptable in Asian cultures (Kagitcibasi, 1996). However, western mothers who breastfeed their children are more likely to adopt bed sharing with their children for the convenience of breastfeeding at night-time (McCoy et al., 2004). These mothers who purposefully sleep with their young children at night are regarded as intentional co-sleepers. Other parents who co-sleep in reaction to the existing sleep problems of their children are classified as reactive co-sleepers. The reactive co-sleepers are more likely to have distress and dissatisfaction because their sleep is also affected (Ramos, 2003; Ward, 2015).

Liu et al., (2006) found that regular co-sleeping was more common among children with ASD and their parents (16% sharing a bed with parents) than among the TD children (5% sharing a bed with parents). Several factors may account for the high frequency of co-sleeping in children with ASD. Firstly, sleep problems are rife in children with ASD. Because of the associated disabilities among children with ASD, mothers may deem their children to be requiring more nurturing during the night. Secondly, restricted and repetitive behaviours which are common in children with ASD might heighten their unwillingness to separate themselves from the parental

bed/room to a new sleep environment. Thirdly, parents possibly want their child to have an undisturbed sleep in order to compensate for the sleep time wasted through their child's reluctance to go to bed (Cotton & Richdale, 2006). Co-sleeping may also be promoted by the extreme reactions of children with ASD towards internal (e.g., unpleasant dreams) and external stimulus (e.g., night-time fears).

### **Aetiology of Sleep Problems in Children with ASD**

The sleep problems in children with ASD are likely to be the result of a combination of biopsychosocial factors (Malow et al., 2006). The bioecological model also highlights a situation that child maladaptive functioning may be a joint effect of the interaction among a child's psychological and biological characteristics, the family, the community and culture. These factors are nested within one another. In the context of sleep of children with ASD, these factors might include social-communication deficits, abnormal melatonin production, comorbid conditions (e.g., anxiety), familial routines (e.g., parent-child interactions at bedtime), and cultural beliefs (Blampied & France, 1993; Richdale, 1999; Richdale & Prior, 1995; Richdale & Schreck, 2009). Some of these are detailed below.

**Social factors.** A relationship between social communication deficits and sleep problems is possible (Richdale, 1999). The sleep-wake cycle is a circadian rhythm that humans use environmental (e.g., daylight) and social cues (e.g., social demands and meal time) to entrain a 24-hour biological rhythm (Johnson, 1996). Routine and social cues are believed to be helpful for young children to establish regular sleep-wake patterns. Children with social-communication deficits may encounter difficulties in using such social cues to entrain their rhythms conducting to irregular and unstable sleep-wake patterns (Baker & Richdale, 2017). For example, a young child might not be able to associate bedtime as always following dinner time.

Thereby, the child does not develop a regular sleep schedule (Mindell & Owens, 2015).

Clinical data of a group of over two thousand children with ASD (aged 4 -18 years) indicated that a child's inability to develop peer relationships was found to be a core symptom in association with the shorter duration of sleep (Veatch et al., 2017). Issues with establishing interpersonal relationships may negatively impact the emotional state of a child and eventually affect the sleep (Ladd & Troop-Gordon, 2003). It is also possible that poor sleep negatively affected the emotional regulation of the children. Poor self-control might prevent them from establishing good interpersonal relationship (Pouw, Rieffe, Stockmann, & Gadow, 2013).

**Biological factors.** Melatonin is a hormone released by the pineal gland to maintain and regulate the circadian sleep-wake cycle and to enhance sleepiness (Armstrong, 1989; Didden & Sigafos, 2001). A person's melatonin level usually starts to increase in the evening once the sun sets, followed by a diminution in the morning upon the sunrise. However, atypical melatonin production such as daytime elevation (Ritvo et al., 1993), decreased amplitude (Nir et al., 1995) and lack of night-time elevation (Kulman et al., 1995) has been found in individuals with ASD. Serotonin, a precursor to melatonin, is also believed to play a crucial role in sleep regulation (Hodge, Carollo, Lewin, Hoffman & Sweeney, 2014). Low levels of serotonin have also been found associated with ASD (Kulman et al., 2000; Nir et al., 1995; Tordjman et al., 2005). Insufficient secretion of melatonin or serotonin may contribute to the difficulty in establishing a 24-hour sleep-wake cycle (Rossignol & Frye, 2011; Tordjman et al., 2005). Abnormalities in melatonin production might contribute to the early-morning waking, frequent night-waking and late sleep-onset of children with ASD (Patzold, Richdale & Tonge, 1998; Richdale & Schreck, 2009).

**Psychological factors.** Anxiety associated with physiological and cognitive arousal could disrupt a resting bodily state and thus adversely affect sleep (El-Sheikh, Buckhalt, Granger & Keller, 2008). Children with ASD were found to have a significantly higher parental rating of anxiety than TD children (Mayes, Calhoun, Murray, Ahuja & Smith, 2011). A positive correlation between sleep difficulties and anxiety symptoms has been found in children with ASD (Rzepecka, McKenzie, McClure, & Murphy, 2011).

Malow et al. (2006) compared the polysomnographic recordings with the data of three psychological domains (i.e., anxious/depressed, affective problems, and attention problems) collected from TD children, good sleepers with ASD (those with no or mild parental sleep concerns) and poor sleepers with ASD (those with severe parental sleep concerns). The ASD good sleepers did not differ from the TD children in sleep architecture or on sleep quality. Poor sleepers with ASD showed more emotional disturbance and fewer social interactions than ASD good sleepers and the TD children. Regarding the anxious/depressed domain, the ASD poor sleepers differed from the TD children (Malow et al., 2006). These findings suggest that emotional problems might negatively affect the sleep quality of children with ASD.

**Family factors.** Besides the complex combination of social, biological, and psychological factors, the family factors may also play an important role in affecting a child's sleep (Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006). Sleep-wake associations are the associations that a child learns to fall asleep by linking one's own sleep with the conditions (e.g., co-sleeping with parents) which are habitually present at the time of sleep onset. By that, during night waking, these same conditions are then required in order to allow the child to fall back into sleep. Parental responses such as nurturing the child's requests might establish reinforcement for their actions,

thus sustaining the sleep problems (Richdale & Wiggs, 2005). Moreover, a family history of marital discord, inconsistent or late bedtimes, parental anxiety and maternal depression predict increased occurrence of sleep problems in children with developmental disabilities (Hiscock & Wake, 2001; Mindell & Owens, 2015; Richdale & Wiggs, 2005). Nevertheless, researchers suggested that when parents are more insistent on implementing early and consistent bedtime routines, the sleep problems in children with ASD are likely to reduce (Carno, Hoffman, Carcillo, & Sanders, 2003).

**Cultural factors.** Cultural differences could produce a difference in sleep-wake habits (Oka, Suzuki, & Inoue, 2008). In Japan, the ‘24-hour society’ lifestyle enables the increased usage of electronic devices after school. Television viewing, video game playing, and computer use at night time have been found to negatively affect the sleep-wake habits of Japanese children (Oka et al., 2008). The percentage of 3-year-old children who sleep later than 22:00 was found to be much higher (52%) in Japan than that in Australia (4.1%; Japanese Society of Child Health, 2001).

Parents from cultures (e.g., Asians) that emphasise the interdependence of family members are more likely to let children share a bedroom or bed with them whereas parents from cultures (e.g., Europeans) that promote independence tend to prefer having their children sleep in a separate bedroom (Kagitcibasi, 1996; McCoy, 2004). Mindell, Sadeh, Wiegand, How and Goh (2010) compared the sleep habits of almost 3,000 children (0 to 36 months) across 16 different cultures and found that young children from predominantly-Asian regions (e.g., Singapore) had significantly later bedtimes and were more likely to share a bedroom with their parents than children from predominantly-European countries (e.g., UK). The figures of bed-sharing with parents ranged from 5.8% (NZ) to 83.2% (Vietnam). Late bedtimes

varied from 19:27 (NZ) to 22:17 (Hong Kong) and total sleep time ranged from 11.6 (Japan) to 13.3 (NZ) hours,  $p < .001$ . However, sharing a room with parents or grandparents was found associated with a lower risk of sleep anxiety in children with ASD (aged 2 to 8 years) in Hong Kong (OR: 0.11, 95% CI 0.012-0.94; Doo & Wing, 2006).

### **Treatment of Sleep Problems in Children with ASD**

Sleep disorders in ASD often remain untreated and ignored (Wiggs & Stores, 1996). It is possible that the ASD associated challenging behaviours always draw more concern and thus take the parents' priority attention and caring over the other problems (Williams, Sears, & Allard, 2004). Moreover, Wiggs and Stores (1996) revealed that less than half of the parents of children with disabilities and sleep disorders received proper sleep treatments. The severity of sleep disturbance in children with ASD has negative concomitant impacts on the parental sleep, the family functioning and the child's daytime behaviours, implying the necessity for effective treatments (Chu & Richdale 2009; Cortesi, Giannotti, Sebastiani, Panunzi, & Valente, 2012; Hodge et al., 2013; Schreck, Mulick & Smith 2004; Taylor, Schreck, & Mulick 2012). Without proper treatment, sleep problems might persist and become worse (Hodge et al., 2014; Wiggs & Stores, 2007). Sleep problems in children with ASD are most commonly treated with behavioural intervention, pharmacological treatment or a combination of both methods. Medication is likely to be the most prevalent way of treatment, although some parents perceive behavioural therapy is a more useful treatment (Cuomo et al., 2017).

**Pharmacological approach.** There is increasing evidence indicating that sleep problems in children with ASD are related to atypical melatonin production (Kulman et al., 2000; Melke et al., 2008; Tordjman et al., 2005). Thereby,



pharmacological interventions, in particular, melatonin, are widely recommended by clinicians as a treatment for sleep problems in children with ASD (Guérolé et al., 2011; Rossignol & Frye, 2011). They have also gained widespread acceptance by parents as it is easy to administer and has an immediate effect (Malow et al., 2012; Schwichtenberg & Malow, 2015). Typically, melatonin is given orally just before the desired sleep onset time. The use of melatonin is found to be efficacious for sleep problems in TD children (Ivanenko Crabtree, Tauman, & Gozal, 2003; Smits et al., 2003) and children with developmental disabilities, including ASD (Cortesi et al., 2012; Niederhofer, Staffen, Pittschieler, & Mair, 2003; Phillips & Appleton, 2004).

For example, Paavonen et al., (2003) reported that the mean sleep latency decreased from 40.02 minutes to 21.82 +/- 9.64 minutes ( $p = .002$ ) after a 14-day melatonin intervention in 15 children with Asperger disorder. In a placebo-controlled study, the researchers included children with ASD and children with Fragile X syndrome. The mean sleep duration increased by 21 minutes ( $p = .02$ ), mean sleep-onset latency reduced by 28 minutes ( $p < .001$ ) and mean sleep-onset time was earlier by 42 minutes ( $p = .02$ ) among those treated with melatonin when compared to the controls (Wirojatan et al., 2009).

Whilst the majority of evidence supports the effectiveness of melatonin, other studies reported limited efficacy of melatonin intervention (Phillips & Appleton, 2004; Schwichtenberg & Malow, 2015). For example, whilst initially effective, the effect of melatonin appeared to fade over time in some children (Rossignol & Frye, 2011; Tordjman et al., 2013). There is also evidence suggesting that melatonin is effective for reducing the sleep onset latency in individuals with ASD, but might not be sufficient for decreasing night wakings (Paavonen et al., 2003). Furthermore, the cessation of melatonin administration often results in a return to the stage of pre-

treatment sleep problems (Doyen et al., 2011; Paavonen et al., 2003). It might imply a long-term pharmacological treatment is needed, which requires more treatment costs and medication compliance. A prolonged melatonin treatment might increase the burden to low SES families. Hock, Kinsman and Ortaglia (2015) examined the situation of treatment adherence among a group of parents of children with ASD and revealed that greater treatment cost was associated with poor adherence to pharmacological treatments. Another shortcoming of medication treatment is that it does not teach children the skills that they need to maintain independent sleep.

**Behavioural approach.** Although melatonin is effective, relatively safe, and well-tolerated in children (Malow, Adkins, et al. 2012), some parents of children with ASD prefer behavioural interventions to pharmacological treatments (Gail et al., 2004). Possible explanations for the parental preference for behavioural interventions include enhancement of the parental sense of competence, control, and ability to cope (Wolfson, Lacks, & Futterman, 1992). Also, behavioural approaches teach children the skills that they need to initiate and maintain sleep.

Some research studies have demonstrated the effectiveness of behavioural interventions in treating the sleep problems of children with ASD (Cortesi et al., 2012; Malow et al. 2014; Reed et al. 2009). Behavioural interventions typically include the modification of antecedents (e.g., inconsistent bedtime) and consequences (e.g., parental attention) that maintain the problem. Functional Behavioural Assessment (FBA) is an evidence-based approach used to inform intervention. In sleep intervention, FBA includes a combination of direct (e.g., observation) and indirect measures (e.g., parent interviews) to identify the antecedents and consequences that have impacts on a child's sleep (Blampied, 2013; Blampied & France, 1993). Treatment plans informed by FBA commonly contain strategies to modify or reduce

the sleep-interfering variables. For instance, parents might use a cuddly toy to replace the child's need to co-sleep with them. Also, a bedtime pass or a card that is exchangeable for a free trip of going out of a child's bedroom could be used to reduce the number of times a child getting out of bed (Galland & Mitchell, 2010).

Examples of antecedent modifications involve the use of sleep hygiene, social story and faded bedtime (McLay, France, Blampied, Danna, & Hunter, 2017; Vriend, Corkum, Moon, & Smith, 2011). The use of sleep hygiene (e.g., consistent bedtime schedule for teeth cleaning and the like) was found effective in reducing night wakings and early awakenings in children with ASD (Durand & Christodulu, 2004; Piazza, Fisher, & Sherer, 1997). The practice of having a regular bedtime was associated with a more than 50% decline in bedtime resistance in children with ASD (Doo & Wings, 2006). Faded bedtime (e.g., systematically delaying the bedtime) paired with the elimination of daytime naps have been found effective in increasing the motivation and need for sleep (Piazza et al., 1997). A social story, a kind of visual aids, tailoring for individual child serves the function of psychoeducation by teaching the child the desirable behaviours about sleep (e.g., putting away toys before bed; McLay et al., 2017).

Consequence-based modifications include strategies such as standard extinction, graduated extinction as well as modified graduated extinction strategies (e.g., faded parental presence and minimal check). Standard extinction is the strategy that requires the parents to ignore all bedtime disruption (e.g., crying out) by withholding any interaction with the child until morning. The parents in the study of Weiskop and colleagues (2001; 2005) adopted this strategy in a group of children with ASD, and it was found that the times of their children's night waking dropped to a lower level and the improvement could maintain at 12-month follow-up stage. The

strategy of faded parental presence, also known as camping out, is a common intervention that refers to the fading away of the presence of a parent in the child's bedroom during bedtime. A parent may sit in a chair next to the child's bed and then gradually moves further away from the child's room. The parent withholds interaction with the child unless for safety issues (France & Blampied, 2005; Honaker & Meltzer, 2014). For minimal check strategy, the parents will respond to a child's awakenings for a while (e.g., give verbal reassurance) and return the child to sleeping position, the parents then leave the room (France & Blampied, 2005; Sadeh, 1994). Research findings also suggested that graduated extinction (i.e., parents ignoring disruptive bedtime behaviours for a predetermined period) could reduce sleep onset latency, co-sleeping and bedtime disturbance in ASD groups and children with insomnia (Blunden, 2011; Durand, Gernett-Dott, & Mapstone, 1996; Moore, 2004).

Despite these benefits, behavioural interventions are not provided to families of children with ASD as frequently as sleep-enhancing medications (Gail et al., 2004). In a randomised placebo-controlled trial, the researchers compared the effectiveness of melatonin treatment alone, cognitive behavioural therapy alone and combined treatment in 140 children with ASD (Cortesi et al., 2012). It should be noted that melatonin therapy alone displayed higher efficacy than cognitive behavioural therapy alone in ameliorating bedtime resistance, sleep onset delay, night-waking and fragmented sleep. Cognitive behavioural therapy alone appeared to be slightly more effective in attenuating sleep anxiety (Cortesi et al., 2012). Most importantly, the group receiving the combination of both types of intervention demonstrated greater improvement in insomnia and with fewer dropouts than the two solo-treatment groups.

**Secondary treatment outcomes.** A family is a unit where the family

members are interdependent on one another, including interactions during the day and night (Bowen, 1985; Bronfenbrenner, 1999). Thereby the improvement of sleep problems in one individual often impacts multiple family members (Dahl & El-Sheikh, 2007). The dynamic and reciprocal interactions among family members imply the mutually beneficial relationship between child sleep and family wellbeing (Meltzer & Montgomery-Downs, 2011). It has been reported that effective treatment helps relieve a child's sleep disturbance resulting in reduced parental stress and enhanced parental mental functioning (Eckerberg, 2004; Scott & Richards, 1990). In one study, Reid, Walter and O'Leary (1999) reported a reduction in the child's sleep problems following behavioural intervention was associated with the psychological stability and parenting competence of the mothers.

The secondary treatment outcomes of sleep intervention include the enhancement of children's daytime behaviour and family functioning. Positive secondary outcomes include improvements in child mood, ASD severity, child social behaviour, child challenging behaviours, mother-child relationship, parental stress, maternal depression, parental sleep, parenting efficacy and marital satisfaction (Durand & Mindell, 1990; France, Blampied, & Wilkinson, 1999; Hiscock & Wake, 2002; Leeson, Barbour, Romaniuk, & Warr, 1994; Sadeh, Gruber, & Raviv, 2002). Therefore, it is imperative to consider a child's sleep problems within a family context (Dahl & El-Sheikh, 2007).

Kazdin and Whitley (2006) suggested that when analysing the behavioural problems of a child, multifaceted case complexity should be considered. Four domains of case complexity, namely, the severity of child dysfunction, socioeconomic disadvantage, parent and family functioning, and barriers encountered during treatment, have been proposed, evaluated and utilised (Kazdin & Whitley, 2006).

## **Chapter 2**

### **Literature Review**

The purpose of this literature review is to gain a deeper understanding of the following aspects; firstly, the impact of family complexity on sleep problems in children with ASD, and secondly, the impact of family complexity on their sleep treatment outcomes. Though Kazdin and Whitley (2006) suggested that case complexity should include both child complexity and family complexity, the focus of the present study was on family complexity. Contextual factors, for example, parental stress, parental psychopathology, marital conflicts, and socioeconomic disadvantage are often associated with the behavioural problems of children (Neece, Green, & Baker, 2012; Shaw, Owens, Vondra, Keenan, & Winslow, 1996; Tomanik, Harris, & Hawkins (2004). Kazdin and Whitley (2006) also operationalised family complexity as SES disadvantage, parent and family factors (e.g., parental stress, depression, psychiatric dysfunction, single-parent family and family relationship) and barriers to treatment participation. These contextual factors may also influence the treatment response of a child over the course of treatment (Kazdin, Holland, & Crowley, 1997; Kazdin & Wassell, 2000).

Interactions within the microsystem, where influences are bi-directional, typically involve interpersonal relationships with siblings, schoolmates and caregivers (Bronfenbrenner & Morris, 2006). How these people interact with the child will affect the development of the child. Similarly, the reaction of the child towards the individuals in the microsystem may also influence how they treat the child in return (Bronfenbrenner & Morris, 2006). In the context of child therapy, supportive interactions will understandably foster the child's growth. Stressful home environment and parental psychopathology might hinder the child's progress. For

instance, Bauminger (2002) evaluated the efficacy of a social skills facilitation program in fifteen high-functioning children with ASD (aged 8 to 17). The results indicated that the involvement and support of members in the microsystem (e.g., parents) had significant contributions to the intervention outcomes. Demands from the exosystem might reduce a family's ability to support a child, that may indirectly affect the child's therapeutic performance. Factors in exosystem such as low SES, stress at work, being a minority group in the community and residing far away from social service providers might limit the parental accessibility to treatment service, and the parental motivation and involvement in child therapy (Algood, Hong, Gourdine, & Williams, 2011; Kazdin, Holland, Crowley, & Breton, 1997; Kazdin, Holland, & Crowley, 1997). As such, contextual factors of family complexity would need to be addressed so as to identify for which child a treatment does work and when the treatment is not enough to help a family (Brestan & Eyberg, 1998; Owens et al., 2003).

Nonetheless, the characteristics of a child could also influence the others and the environment, that eventually could have an impact on the treatment outcomes. Bronfenbrenner and Morris (1998) suggested that the physical (e.g., age and gender), mental (e.g., cognitive ability and skills) and emotional characteristics (e.g., temperament and motivation) of a child are the resources that a child brings to the social situation in the environment. These resources might have an impact on the environment and others, such as the intensity of treatment, the caring of parents and the attention of therapists. By that, the child can influence the treatment outcomes.

The focus of this review is on how these family factors might impact on the sleep problems and treatment outcomes of the children with ASD. However, the direction of any causal relationship between the child sleep and parental functioning is

inconclusive. Therefore, this literature review also presents a brief review of how the child factors might affect the family functioning as well as the treatment outcomes so as to illustrate the interconnectedness between child sleep and family factors in the ecological system.

In this review, a brief review of child factors on treatment outcomes, and child sleep on family functioning are presented first. After that, the impact of familial factors on child sleep and treatment outcomes is discussed based on the contextual framework of parent/family factors proposed by Kazdin and Whitney (2006), such as parental stress, parental depressive symptoms and marital relationship. For the purpose of the present study, parental sleep quality was also included in this review. Literature pertaining to the familial stressors or obstacles such as low SES, large family size, parental psychopathology, parental criminality, siblings having developmental difficulties, parent-therapist communication difficulties, disagreement in parents over child-rearing, geographical obstacles, and other possible barriers that might interfere with treatment participation are also reviewed. The limitations of the past studies are summarised to denote the areas that require more research evidence.

### **Search process**

Electronic databases (e.g., Google Scholar, PsychINFO and Science Direct) were used to identify relevant articles. Examples of search terms included, 'sleep\*', 'ASD/autis\*', 'Pervasive Developmental Disorder', 'PDD', 'developmental dis\*', 'DD', 'family\*', 'parent\*', 'child\*', 'adolescen\*', 'family', 'parent\*', 'parent\* wellbeing', 'marital relationship', 'maternal', 'complexity', 'family characteristics', 'family factors', 'treatment participation', 'therapeutic outcome', 'communication barrier', 'treatment acceptability', 'treatment adherence', 'behavioural intervention', 'cognitive behavioural therapy', 'CBT', 'treatment' and 'treatment outcome', 'family



adversity’, ‘risk factor’, ‘predictor’, ‘socio\* disadvantage’, ‘family stressors’, ‘barrier’, ‘obstacles’, etc. The reference lists of systematic reviews and meta-analyses were also checked for relevant articles. The initial hit yielded several thousand articles depending on the combination of keywords used in searching. Studies that were unavailable in English, were not focused on children, were used with interventions other than behavioural intervention or CBT, were not focused on sleep difficulties or behavioural problems, and did not include family characteristics as correlates or predictors of child sleep problems were excluded. To increase the number of available studies, case study, studies with small sample sizes, and non-RCTs were also reviewed. Studies that recruit children with developmental disabilities other than ASD were also included.

### **Child Factors Related to Treatment Outcomes**

Age of the child, ASD severity, pre-treatment functioning level during the treatment commencement are the possible predictors of treatment outcomes of children with ASD (Perry et al., 2011; Virues-Ortega, Rodríguez, & Yu, 2013). The younger the brain, the more the malleability it has, implying that younger children might benefit more from behavioural interventions than their older counterpart (Matson, Wilkins, & Macken, 2008). The belief of ‘the earlier the age of entry to treatment the better’ has received empirical evidence from studies that investigated the outcome of behavioural therapy in children with ASD (Anderson, Avery, DiPietro, Edwards, & Christian, 1987; Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009; Harris & Handleman, 2000; Perry et al., 2011).

Although research commonly suggests early intervention appears to be more effective for young children, some variables are found also related to the different pace of progress at post-treatment (i.e., some children gain more and some gain less).

Pre-treatment cognitive functioning, social and adaptive functioning, autism severity and language ability could also be predictors of treatment response (Perry et al., 2011; Virues-Ortega, Rodríguez, & Yu, 2013; Zachor & Itzhak, 2010). For example, Sallows and Graupner (2005) reported that children with low autism severity (together with better cognitive abilities) at treatment onset were more likely to have more intervention gains. However, Remington et al. (2007) found that those with higher autism severity achieved more progress at the post-intervention stage.

The few studies that include demographic factors have found that gender or ethnicity are not associated with treatment outcomes in ASD (Baker-Ericzén, Stahmer, & Burns, 2007; Mandell et al., 2013).

**Impact of children's sleep problem on the family.** The sleep difficulties of children with ASD often place an additional burden on their parents (Honomichl, Goodlin-Jones, Burnham, Gaylor, & Anders, 2002; Johnson 1996, Richdale 1999). Parental stress, parental psychological health, parental sleep and marital relationship have been found being negatively affected by the sleep problems in children with ASD (Hodge et al., 2013; Reid et al., 1999; Richdale et al., 2000).

For example, Hodge and colleagues (2013) studied a sample of 90 parents and their children with ASD. The findings suggested two possible pathways that children's sleep problems could influence the wellbeing of their parents. Firstly, children's sleep difficulties could elevate the parental stress levels leading to disruption to parental mental wellbeing (Hodge et al., 2013; Richdale et al., 2000). Richdale and colleagues (2000) investigated the sleep problems of 52 children (ages 2 to 19) with intellectual disability (including seven children with ASD) and discovered that sleep problems were related to both the intensity and frequency of child resistance (e.g., settling difficulties and yelling) that could bring considerable stress to their

parents. The survey data collected from 210 parents of children with pervasive development disorder (mean age 3.5 years old) revealed that the parents of children with disrupted sleep experienced a higher level of stress than those whose children showed no sleep problem (Doo & Wing, 2006).

Secondly, in children with developmental disabilities, children's sleep habits have been found reducing the sleep quality of their parents, which might also eventually predict mental health problems in the parents (Chu & Richdale, 2009). Recent estimates suggested that over 60% of parents of children with ASD had disrupted sleep due to their child's sleep difficulties (Polimeni et al., 2005). The actigraphic recordings revealed that parents of children with ASD had an earlier wake time and shorter sleep duration compared with parents of TD children (Meltzer, 2008).

Thirdly, studies in TD children suggested that emotional and behavioural problems associated with children's sleep problems could lead to stressful family dynamics which would incur pressure on the marital relationship (El-Sheikh, Buckhalt, Mize, & Acebo, 2006; El-Sheikh & Dahl, 2007; El-Sheikh & Kelly, 2011; Meltzer & Westin, 2011). It is also reported that parents of children with ASD have higher rates of divorce than parents of TD children (23.5% vs 13.8%) (Hartley et al., 2010).

Finally, the direction of any causal relationship between the disrupted sleep of children with ASD and the parental mental wellbeing is inconclusive. There could be a bi-directional or circular interaction between child sleep and familial contextual factors (Hodge et al., 2013; Lam, Hisock, & Wake, 2003). Despite the increasing studies that focus on the impact of children's sleep problems on parental wellbeing, only a few studies have researched into the impact of family complexity on children's

sleep problems.

### **Family Complexity and Treatment Outcomes**

Efforts to enhance therapeutic outcome have been increasingly focused on family dynamics that may be responsible for the trajectory of disorder, development of sleep problems, and outcome of treatment. Outcome prediction is essential for treatment planning and prognosis. Caregivers play a critical role in a child's behavioural change, particularly in the context of child sleep therapy, where interventions are commonly implemented with parental involvement. The efficacy of behavioural intervention relies on the parents to implement the parenting strategies to teach the child new ways of adaptation (Snyder & Lopez, 2009). In an early behavioural intervention programme provided to children with ASD, pre-treatment functioning of the children (e.g., cognitive ability) appeared to be a possible predictor of treatment gains in the first stage of therapy. However, intervention intensity (i.e., weekly hours) and intervention duration (i.e., total weeks of intervention) were found to be the two main predictors of longitudinal growth in these children regardless of their pre-treatment functioning and age (Virues-Ortega & Rodriguez, 2013). Therefore, a supportive home environment, parental compliance to treatment and good parental functioning might be necessary for a child to retain in an enduring treatment process (Kazdin & Weisz, 1998; Virues-Ortega & Rodriguez, 2013). Bronfenbrenner and Ceci (1994) also emphasised that genetic potentials (i.e., heritability) alone do not yield 'finished traits' (p.572). A child's development is the product of the genetic potential and the environment which involved the enduring parent-child activities in the process (Bronfenbrenner, 1995).

A growing number of studies has explored the family factors that could have impact on the treatment outcomes in TD children with behavioural problems

(Armbruster & Kazdin, 1994; Harland, Reijneveld, Brugman, Verloove-Vanhorick, & Verhulst, 2002; Kazdin et al., 1997; Kazdin & Wassell, 2000; Maughan, 2001; Redline et al., 1999; Reid, Hong & Wade, 2009; Robbins, Dunlap & Plenis, 1991). The researchers reported that low SES (e.g., unemployment), being an ethnic minority, parental stress, family dysfunction (e.g., marital conflict, domestic violence) and difficult living circumstances (e.g., single-parenthood) might be the salient factors that predict slow progress or drop-out in child psychotherapy (Kazdin et al., 1997; Kazdin & Wassell, 2000; Maughan, 2001; Redline et al., 1999; Reid et al., 2009; Robbins et al., 1991). Adverse life events and parental psychopathology were also found to be barriers to treatment participation of a family (Armbruster & Kazdin, 1994).

Lam et al., (2003) reported that children from families with high adversity might be more likely to have persistent and recurrent sleep problems. Studies have revealed an array of family factors that may serve to complicate the trajectory of ASD and the associated problems (Fountain, Winter, & Bearman, 2012; Lucia & Breslau, 2006; Midouhas, Yogaratnam, Flouri, & Charman, 2013). Families of children with ASD often experience increased relationship conflict, maternal psychopathology, disrupted sleep in siblings and parents, and family dysfunction (Lopez-Wagner, Hoffman, Sweeney, Hodge, & Gilliam, 2008; Mauldon, 1992; Midouhas et al., 2013; Polimeni et al., 2005; Wiggs & Stores, 2004). Thereby, the developmental outcome of children with ASD may also be influenced by these complex family factors (Fountain, Winter, & Bearman, 2012; Lucia & Breslau, 2006; Midouhas et al., 2013). Nevertheless, there has been little systematic investigation into how family complexities relate to treatment outcomes in children with ASD (Howlin et al., 2009). Family variables are seldom considered as predictors in outcome studies in ASD

context (Vivanti, Prior, Williams, & Dissanayake, 2014).

**Impact of parental stress on children's sleep problems.** Parents of children with ASD are particularly worried about the future of their children because of their cognitive deficits and inability to live independently, as well as the social acceptance of their children (Koegel et al. 1992). Moreover, it has been estimated that approximately 94% of children with ASD demonstrate at least one form of challenging behaviours (Matson et al., 2008). Children with ASD have been reported engaging in higher levels of challenging behaviour than TD children ( $p < .001$ ; Matson et al., 2008). The exacerbated parenting stress in families having children with ASD are predictable.

However, the relationships between children's sleep problems and parental stress are complicated. Parental stress can be a contributor to or a consequence of a child's sleep problem. The burden of taking care of a child who sleeps badly and wakes up frequently during the night may be conducive to chronic sleep interruption in parents that may increase the parental stress level. It is also possible that the stressed parents unintentionally sustain the children's sleep problems by indulging them more (Quine, 1991). Consistent bedtime routine, minimised distractions (e.g., putting away toys) and suitable bedroom environment (e.g., dark without excessive noise) would create conditions that facilitate a child's sleep (Adams & Rickert, 1989; Dahl & El-Sheikh, 2007; Spruyt et al., 2005). Parental stress may reduce the caregiving capacity of the parents, which makes the parents more difficult to maintain an appropriate bedtime arrangement for their children. Because of that, the children might have persistent and recurrent sleep problems.

**Impact of parental stress on treatment outcomes.** Studies that focus on how parental stress might affect the therapeutic sleep outcome in ASD are scarce. A study

conducted by Robbins, Dunlap and Plenis (1991) was possibly the first study focusing on the influence of family factors on therapeutic outcomes in ASD early learning interventions. Findings suggested that maternal stress level showed a highly significant and inverse relationship with the amount of child's progress in the family-oriented training programme. The findings of a retrospective study have revealed that family stress factors (e.g. financial strain and lack of social support) are possible predictors of therapeutic outcomes in children with ASD (Gabriels, Hill, Pierce, Rogers, & Wehner, 2001).

**Impact of parental sleep on children's sleep problems.** Fatigue is a predictable consequence of poor sleep. Parental fatigue was found to be significantly associated with poor parental self-efficacy, low parenting warmth and involvement, and high parenting hostility (Giallo, Rose, & Vittorino, 2011). It might be difficult for a tired or exhausted parent to appropriately implement consistent bedtime routine every night. Thereby, a fatigued parent might be less able to provide the appropriate structure and consequences when the child shows bedtime refusal behaviours. These parenting behaviours might exacerbate the child's sleep problems.

The nature of the relationship between parental sleep and children's sleep problems is not conclusive. Importantly, research evidence suggests that the improvement in children's sleep can positively affect the parental sleep quality (Durand & Mindell, 1990; France, Blampied, & Wilkinson, 1999; Hiscock & Wake, 2002; Leeson, Barbour, Romaniuk, & Warr, 1994; Sadeh et al., 2002; Wiggs & Stores, 2001). For example, Wiggs and Stores (2001) investigated parental sleep, daytime sleepiness and parental perceived ability to control the child's difficult sleep-related behaviour in a group of school-age children with severe intellectual disabilities. The findings revealed that, following the child sleep intervention, an

improvement in the children's sleep quality could predict parental satisfaction in their own sleep as well as in their child's sleep. The maternal daytime sleepiness reduced, and maternal confidence in coping with the child's sleep problems increased. As such, enhanced parental mental wellbeing could also be anticipated.

A large majority of child sleep studies have focused on investigating the impact of sleep problems in children with developmental disabilities on parental sleep quality (Chu & Richdale, 2009; Lopez-Wagner et al., 2008; Polimeni, Richdale & Francis, 2005). Few studies have explored the impact of parental sleep on sleep problems of children with ASD (Vivanti et al., 2014).

**Impact of parental sleep on treatment outcomes.** In comparison to the number of studies that focus on impact of child sleep on parental sleep, far less research effort has been invested in examining the impact of parental sleep on children's treatment outcomes in ASD context (Vivanti et al., 2014). More importantly, fathers' sleep quality is particularly lacking research attention (Hall, Moynihan, Bhagat, & Wooldridge, 2017). Parental stress was found related to the decrease in treatment effectiveness in TD children who had been referred for aggressive and antisocial behaviour therapy (Kazdin & Wassell, 2000). Poor sleep quality might be a consequence of stress. As such, sleep deficiency in parents might reduce the parents' ability to implement strategies in maintaining good sleep habits for their children, which might limit the treatment efficacy or lead to premature withdrawal from the therapy (Kazdin & Wassell, 2000). However, there is a lack of sleep studies investigating the effect of parental sleep on sleep treatment outcomes in ASD context.

**Impact of parental relationship on children's sleep problems.** Family conflict was found to be more predictive of ASD symptom severity than positive



family influences (Kelly, Garnett, Attwood, & Peterson, 2008). To date, there has been very little systematic investigation into the impact of marital conflicts on child sleep in children with ASD, and the directionality of this relationship. It is important to note that, just as children's sleep problems can influence the marital relationship, marital conflict can disrupt the neurobiological regulation (i.e., sleep) of a child (Meltzer & Westin, 2011). For example, in a 2-year longitudinal study, Kelly and El-Sheikh (2011) found that there was a reciprocal relationship between marital conflict and children's sleep disruptions, suggesting exposure to marital conflicts might predict an increase in children's sleep problems over time.

Exposure to familial stressors might increase a child's sensitivity or reactivity to environmental changes (e.g., mood swings in parents due to marital conflicts) and eventually lead to maladaptive biological regulation as indexed by increased sleep disturbance (El-Sheikh, Buckhalt, Cummings, & Keller, 2007). Marital discord may be one of the home stressors affecting the sleep of a child. Current evidence indicates that familial stressors can impair a child's sleep as manifested by extended sleep onset latency and increased night waking (Sadeh, 1996). El-Sheikh et al. (2006) studied a group of TD children (mean age = 8.85 years) discovering that marital discord, especially child-reported conflict, was predictive of reduced sleep duration, elevated sleep fragmentation and increased subjective sleepiness in these children. The sensitisation hypothesis proposes that exposure to inter-parental conflicts over time is predictive of greater child emotional and behavioural reactivity to the external stimulus (Cummings & Davies, 2002). Empirical evidence has indicated that chronic exposure to family conflicts could affect a child's physiological functioning such as vagal suppression (El-Sheikh, Harger, & Whitson, 2001) and electrodermal response (El-Sheikh, 2005), predicting dysfunctions at bio-behavioural functioning (e.g.,

anxiety and hypersensitivity). Feelings of hypervigilance, insecurity, and anxiety might disrupt a child's sleep (Cummings & Davies, 2002). However, there is a scant amount of literature on the effects of marital conflict on the sleep of children with ASD.

**Impact of parental relationship on treatment outcomes.** Research evidence suggests that marital relationships and relationship satisfaction may have impacts on therapeutic sleep efficacy. For example, Carpenter (1990) studied the effectiveness of behavioural therapy for TD children with sleep disturbance. In this study, although 73% of the parents reported improvement in children's sleep, findings from those displayed no improvement suggested that marital problems might account for the limited treatment efficacy (Carpenter, 1990). Similarly, Jones and Verduyn (1983) found that a behavioural therapy successfully reduced the sleep disturbance of TD children, yet the response to treatment was limited when marital conflicts existed in the family. Studies that have investigated the relationship between marital satisfaction and treatment outcomes in children with ASD are rare; further research is required in this area. A greater understanding of relationship nature between marital discord and treatment outcomes may assist clinicians in their work with families by supporting them to identify potential barriers to treatment and to prioritise treatments.

### **Other familial stressors**

Besides the factors related to parental functioning, other environmental factors might also affect the treatment outcomes in ASD context (Vivanti et al., 2014). Bronfenbrenner (1992) explained that ecological niches are “particular regions in the environment that are particularly favourable or unfavourable to the development of individuals with specific personal characteristics” (p. 194). Bronfenbrenner (1995) regarded that when proximal processes occur within unfavourable environments, the

effectiveness of child-adult activities would be reduced. On the other hand, Rutter et al., (1975) and Rutter and Quinton (1977) identified the adversity factors (e.g., low family SES or large family size) in the environment of a child that might increase the risk of maladaptive behaviours in children. Kazdin and co-workers (1997) also proposed that disagreement between parents on treatment goals, geographical barriers and poor parent-therapist relationship could have negative influences on treatment response of children with disruptive behaviour. Although familial factors are rarely treated as predictors in outcome studies in ASD context, the relevance of these adversity factors on child sleep might help clinicians identify the potential barriers to sleep intervention in children with ASD.

**Family socioeconomic status.** It has been reported that sociodemographic disadvantages are related to family dysfunction and maternal depressive symptoms, whereas family dysfunction and maternal psychopathology both predict negative parenting which subsequently leads to children's sleep problems (Reid et al., 2009). Economic obstacle for transportation can be a barrier for low-income families to access the health care service. As such, a delay or interruption to the treatment might intensify and prolong a child's behavioural problems (Kazdin et al., 1997). The absence of risk factors may serve as a protective factor which might reduce the problem severity of a child as well as the chance of premature treatment termination (Kazdin et al., 1997). Doo and Wing (2006) found that the situation of having a working mother (i.e., absence of unemployment) was associated with a lower risk of sleep anxiety in children with ASD.

In contrast, low family income was found to have no influence on therapeutic outcomes in children with oppositional, aggressive and antisocial behaviour (Kazdin et al., 1997). Rather, the cumulative effect of some family factors appears to increase

the chance of premature treatment termination (Kazdin et al., 1997).

In ASD studies, Chervin et al., (2003) also reported that low SES was not associated with sleep problems in children. A large-scale study which recruited 4,470 families found that higher SES would shorten the total sleep time of both children and parents. The children's advancing wake-up time and their delayed bedtime might be related to the increasing use of media and homework load (Zhang, Li, Fong, & Wing, 2010). Nevertheless, a retrospective study reported that low family income could be a possible predictor of low treatment response in children with ASD (Gabriels et al., 2001).

**Family size.** Increased number of children in a family might reduce the time that the parents can spend on each child. The parents might also find it difficult to attend to a child's individual needs (Sadeh & Anders, 1993). A regular sleep routine requires the parental investment of time to implement. Each child might have different needs and thus require a different sleep regime. For instance, a child with ASD might need more guidance before bedtime than a TD sibling. An increased number of siblings might diminish the parents' capacity for maintaining the sleep routines of the child with ASD, which might sustain the sleep problems of all the children. Chou et al. (2012) have reported that maternal overprotection and inability in keeping sleep schedules are associated with the sleep problems of the children with ASD and their siblings. The findings indicated that children with ASD had more sleep problems, including insomnia, sleep-wake cycle disorders and daytime sleepiness. Their non-autistic siblings also had a higher risk of having insomnia, sleep-talking and nightmares, compared to the TD children (Chou et al., 2012).

**Parental psychopathology.** Research has revealed that parental psychopathology (e.g., parental depression or parental substance abuse) is one of the

barriers to optimal treatment response for children with attention deficit hyperactivity disorder (ADHD; Chronis, Chacko, Fabiano, Wymbs, & Pelham, 2004), children with conduct disorder (Webster-Stratton, 1990), and children with externalising behaviours (Baydar, Reid, & Webster-Stratton, 2003; Fuller et al., 2003). Moreover, parental psychopathology was found to be correlated with high school drop-out rates and poor compliance with behavioural interventions (McMahon et al., 1981). Nevertheless, the role of parental psychopathology in ASD treatment outcomes, with a particular focus on sleep, has received no attention so far.

**Parental criminality.** Parental criminality might reduce parental involvement in child-rearing (Barber, 2000). Neglectful parental behaviour and lack of emotional support from parents who engaged in criminal acts or in imprisonment might increase the risk of developing maladaptive behaviour in children (Barber, 2000). Absence of fathers has been found correlated with poor sleep in TD children (Bell & Belsky, 2008; Bernier, Bélanger, Bordeleau, & Carrier, 2013). However, parental criminality has never been examined in sleep studies in children with ASD.

**Sibling(s) having developmental disorders.** The studies of family and twins have revealed that ASD is highly heritable within a family, with some estimating that the heritability could be as high as 95% (Bailey et al., 1995; Rosenberg et al., 2009; Szatmari, Jones, Zwaigenbaum, & MacLean, 1998). The prevalence of an ASD phenotype in siblings of children with ASD has also been reported to be 40% (Losh, Childress, Lam, & Piven, 2008). The high demand of time in taking care of several children with special needs has been identified as one of the situational barriers to child behavioural interventions (Koerting et al., 2013).

**Parent-therapist communication barriers.** The studies of Kazdin and colleagues (1997; 2006) are possibly the very few studies that examined the barriers

experienced by families during treatment. However, these studies were conducted in the context of child outpatient psychiatric treatment. There seem to be no studies that have examined the parent-therapist communication barriers in the child sleep literature.

In working with families, the family system perspective emphasises that parents and professionals are partners (Cridland et al., 2014). Key characteristics of effective parent-therapist partnership in children behavioural therapy include mutual agreement on therapeutic goals, shared planning and decision-making (Morrow & Malin, 2004). Inadequate communication could lead to misunderstanding and a disrupted working relationship. Communication difficulties between parents and therapists could be attributed to language barriers, cultural difference, inadequate understanding or misconception about the interventions and insufficient confidence and trust in the professionals (Keen, 2007).

Treatment acceptability refers to the extent to which the clients perceive the treatment as acceptable, reasonable, and attractive (Kazdin, 2000). Elliott (1988) suggested that treatment acceptability is a crucial factor in determining the success of a treatment plan, particularly at the initial stages of the treatment process. High acceptability of a treatment plan is likely to enhance a client's adherence to the treatment procedures (Elliott, 1988). Treatment adherence may optimise the effectiveness of an intervention (Moore & Symons, 2009). Prinz and Miller (1994) have reported that those who prematurely drop out from mental health services are less likely to show gains from treatment than those who remain in treatment.

To date, very limited studies have investigated the relationship between parental ratings of treatment acceptability and treatment outcomes. However, evidence suggests that, during the intervention phase, the barriers that parents have

encountered and the improvement that children have demonstrated may predict treatment acceptability ratings in children with antisocial behaviour (Kazdin, 2000). In one study, Weiskop, Richdale and Matthews (2005) provided sleep intervention for a group of 13 children with ASD or Fragile X syndrome (aged 3.5 to 9 years). Their parents reported that they appreciated the parent-therapist communication (e.g., phone calls), the support from the therapists, the information about goal-setting and the child's progress report provided by the researchers. However, the mothers also identified that record keeping, adherence to the bedtime routine, and the time-consuming nature of the programme were the obstacles they experienced. Developing a better understanding of how the parents perceive the treatment may help clinicians design treatment with better social validity.

**Disagreement in parents over child rearing.** Parent training studies have revealed that when teaching the parents to modify their children's problem behaviour, the parental failure to cooperate in treatment sessions has been identified as one of the barriers to effective treatment (Forehand, Middlebrook, Rogers, & Steffe, 1983). Although parents would work together in child rearing, they may have different expectations or strategies on how to handle the problems of their children, which can be a possible source of distress (Knafl & Deatrick, 2003). Johnson, Frenn, Feetham, & Simpson (2011) studied the parents of children with ASD and reported that mothers and fathers were likely to have different expectations in child rearing, caregiving, and family functioning. A higher level of discrepancy between two parents' perceptions and expectations in family functioning was associated with lower parental wellbeing (Johnson et al., 2011).

**Geographical obstacles.** Geographical distance could be a challenge to families with children with disabilities for accessing health care service. In a review,

Keorting et al. (2013) revealed that practical issues such as transport cost and inconvenient timing or venue could be the possible barriers to interventions for child behaviour problems. This is particularly apparent in rural and remote areas, where communities are widely dispersed and access to health care service is limited (Smith & Gray, 2009). Long distance travel and lack of transportation to reach the health service providers are the possible barriers to treatment participation (Ashburner, Vickerstaff, Beetge, & Copley, 2016).

**Other barriers to treatment participation.** Parental attendance and adherence are essential features of child-centred therapy (Kazdin, Holland, Crowley et al., 1997; Nock & Ferriter, 2005). Treatment attendance refers to the presence of participants in the treatment appointments or training sessions (Nock & Ferriter, 2005). Treatment adherence indicates the willingness or ability to follow the intervention procedures or strategies (Meichenbaum & Turk, 1987). In a review, Nock and Ferriter (2005) have reported that failing to attend treatment sessions or reluctance to comply to treatment procedures are associated with negative clinical outcomes or poor treatment participation. Although most child-centred interventions require high levels of parental adherence and attendance, the evaluation of these family factors in behavioural treatment research is surprisingly limited (Chamberlain, Patterson, Reid, Kavanagh, & Forgatch, 1984; Nock & Ferriter, 2005). These family factors are also not examined in sleep studies in ASD context.

### **Limitation of Past Studies**

There is very limited systematic investigation into the relationship between parent or family characteristics and treatment outcomes in child sleep (Mindell et al., 2006). Sadeh and Anders (1993) reviewed the family factors that might perpetuate the sleep problems of TD infants. The factors that they identified included parent working



hours, family size, and socioeconomic status (Sadeh & Anders, 1993). Socioeconomic concerns such as unstable employment that undermines the economic stability of a family and expands the irregularity of day-night schedules could put an additional challenge on the process of resolving the sleep problems of children (Sadeh & Anders, 1993). It has been suggested that familial demographics such as maternal education level, maternal age, parental relationship status (partnered versus solo-parented), and family size are the potential predictors of treatment outcomes in children with conduct problems (Beauchaine, Webster-Stratton, & Reid, 2005). Although these papers revealed some valuable findings regarding the sleep of children, little is known about the family factors that might be associated with sleep treatment outcomes in children and adolescents with ASD.

Despite this emerging literature, existing research on family predictors of treatment outcomes has examined a relatively narrow set of family features and produced an inconclusive pattern of findings. Most researchers have examined the impact of only one or two characteristics of family complexity (e.g., SES and marital relationship) on children's sleep. When more characteristics are studied together in one study, no single characteristic seems to be necessary or sufficient to affect the treatment outcomes in behavioural treatment (Kazdin & Mazurick 1994). This might imply that there is an intertwining effect among the different family characteristics which produce a synergistic impact on the treatment outcomes. Thereby, a complexly interacting effect might present underneath the dynamics of a family. So far, no research with a particular focus on sleep problems has predicted the sleep treatment outcomes in children with ASD by family complexity.

Whilst there is a large corpus of evidence to support the effectiveness of behavioural sleep intervention (Cuomo et al., 2017; Rigney et al., 2018), most of these

studies are limited to case studies or studies with a small sample size ( $n \leq 10$ ; e.g., Durand et al., 1996; Christodulu & Durand, 2004; Weiskop et al., 2005). A small sample size with less than 10 participants might limit the ability of a study to correctly reject the null hypothesis (VanVoorhis & Morgan, 2007). Pellechia et al. (2016) also commented that most behavioural studies in the ASD context had a small sample size making these studies underpowered for studying the predictors of outcome. As a result, it is difficult to generalise research findings across the ASD population. The importance of conducting a study with a larger sample size is therefore underlined.

### **Rationale of the Present Study**

There is evidence suggesting that sleep problems in children with ASD have a profound impact on parental wellbeing, parental sleep quality, and family functioning and relationships. There are, however, few studies that have investigated the impact of family complexity on sleep problem severity and treatment outcomes among children with ASD. Although there is very limited research in this area, a few researchers suggest that family adversity, parental wellbeing, marital satisfaction, maternal psychopathology, and the number of children in a family might be associated with the outcome of sleep intervention in children with ASD (Doo and Wing 2006; Kelly et al., 2008; Robbins et al. 1991). Moreover, although treatment acceptability is viewed as an important consideration in intervention research (Gresham & Lopez, 1996) and applied behaviour analysis (Schwartz & Baer, 1991), the association between treatment outcomes and treatment acceptability ratings is not clear. The present study might guide the development of appropriate treatments aimed to improve sleep problems in children with ASD.

The present study aimed at investigating (1) whether parent-implemented behavioural sleep treatment has altered the severity of sleep problem in children with

ASD, (2) whether there was any complexity variable residing underneath a cluster of parent and family variables obtained at pre-treatment, (3) whether the components of family complexity prior to treatment could predict the treatment outcomes, and (4) whether ratings of treatment satisfaction were related to treatment response of the children. Moreover, in compared to previous studies, the present study with a bigger sample size might enable the possible correlations or effects to manifest more clearly.

### **Research Questions**

The present study investigated the following research questions:

1. Did the treatment alter the child's sleep to a clinically significant degree?
2. Was there any complexity component laying underneath the cluster of multiple parent/family variables measured at pre-treatment?
3. Did the family complexity components and any other family variables measured at pre-treatment predict the treatment outcomes?
4. Was there any correlation between treatment outcomes and parental ratings of treatment acceptability?

## **Chapter 3**

### **Method**

The present study was inspired by the framework proposed by Kazdin and Whitley (2006) who noted that case complexity could be measured in terms of child functioning characteristics, parent and family factors, and barriers to treatment participation. The focus of the present study was to investigate whether parent and family factors would have an impact on the treatment outcomes in children with ASD. To achieve this, the components that resided underneath multiple parent and family variables obtained at pre-treatment were explored. The present research focused on the analysis of both questionnaire and clinical interview data gathered pre- and post-intervention. The data set used in the current study was extracted from the data pool of a larger New Zealand-wide sleep and autism study that recruited children with ASD who were experiencing sleep problems. Data on child and family characteristics were gathered during pre-treatment and assessment phases of the larger study using a combination of clinical interviewing, sleep assessment measures, sleep diaries, video-recording, psychometrics of wellbeing and conference discussion with the psychologists responsible for the families. Once collected, data were used in the present study to investigate the relationship between family complexity and children's treatment outcomes, in particular how well the components of family complexity (i.e., independent variables) could predict the children's amelioration of sleep problem severity (i.e., dependent variable) in sleep intervention. This chapter describes the present study within the framework of the larger sleep focusing on sleep problems of children with ASD.

## **Operational Definitions and Variables**

**Family complexity.** For the purpose of the present study, the following variables the family demonstrated at pre-treatment assessment phase were used to define family complexity operationally. The variables included: (1) the parental mood states, as measured using the Depression Anxiety Stress Scale-21 (DASS-21; S.H. Lovibond & P.F. Lovibond, 1995), (2) the parental sleep quality, as measured on the Pittsburgh Sleep Quality Index (PSQI; Buysse, Reynolds, Monk, Berman, & Kupfer, 1989), (3) marital satisfaction, as measured on the Relationship Quality Index (RQI; Norton, 1983), and (4) a composite family adversity score that addressed family sociodemographic factors and barriers to treatment participation. So that, 11 parent/family variables obtained at pre-treatment were used to define family complexity, namely, (1) maternal DASS-depression (DASS-D) pre-treatment, (2) maternal DASS-anxiety (DASS-A) pre-treatment, (3) maternal DASS-stress (DASS-S) pre-treatment, (4) paternal DASS-depression pre-treatment, (5) paternal DASS-anxiety pre-treatment, (6) paternal DASS-stress pre-treatment, (7) maternal PSQI pre-treatment, (8) paternal PSQI pre-treatment, (9) maternal RQI pre-treatment, (10) paternal RQI pre-treatment, and (11) family adversity score. A summary of the variables included in the present study for defining family complexity was presented in Table 1.

**Children sleep problem severity.** The operational definition of the sleep problem severity in the present study was the sleep problem severity (SPS) score. The SPS score is a composite score that considers the intensity of sleep problems of a child, for example, the duration of sleep each night and the frequency of co-sleeping. In the present study, there were four outcome variables, namely, SPS at baseline (SPS BL), SPS at post-treatment (SPS Post-tx), SPS at short-term follow-up (SPS STFU)

and SPS at long-term follow-up (SPS LTFU).

**Treatment outcome.** To quantify the treatment outcome, the operational definition of treatment outcome (i.e., the predicted variable) was defined as the change in SPS scores between two time-points. As a result, there were three SPS change scores. They were the change SPS score between (1) baseline and post-treatment (SPS BL vs SPS Post-tx), (2) baseline and short-term follow-up (SPS BL vs SPS STFU), and (3) baseline and long-term follow-up (SPS BL vs SPS LTFU).

**Independent and dependent variables.** Based on the results of four factor analysis models in the present study, the three components that resided underneath the 11 pre-treatment parent/family variables of family complexity found were, (1) maternal wellbeing that included maternal depression, anxiety, stress and sleep quality obtained at pre-treatment, (2) paternal wellbeing that consisted of paternal depression, anxiety, stress and sleep quality obtained at pre-treatment, and (3) parental relationship quality that contained maternal marital satisfaction and paternal marital satisfaction assessed at pre-treatment. The family adversity variable was included in maternal wellbeing component in two of the four factor models. Details of factor analysis models are explained later in the result section. The three components were treated as independent variables (or predictors) to predict the treatment outcomes (see Table 1). The three SPS change scores (i.e., treatment outcomes) were treated as the dependent variables to be predicted by the three components of family complexity.

Table 1

*Summary of Parent/Family Variables for Defining Family Complexity*

Family complexity	
Components resulted from factor analysis (Independent variables)	Parent/family variables included in each component
Maternal wellbeing	Maternal DASS-depression pre-treatment
	Maternal DASS-anxiety pre-treatment
	Maternal DASS-stress pre-treatment
	Maternal PSQI pre-treatment
	Family adversity score <sup>a</sup>
Paternal wellbeing	Paternal DASS-depression pre-treatment
	Paternal DASS-anxiety pre-treatment
	Paternal DASS-stress pre-treatment
	Paternal PSQI pre-treatment
Parental relationship	Maternal RQI pre-treatment
	Paternal RQI pre-treatment

*Note:* The components extracted from factor analysis were the underlying domains that resided underneath the 11 pre-treatment parent/family variables measured for defining family complexity; <sup>a</sup> family adversity were included in maternal wellbeing components in two of the four factor models only.

## **An Overview of the Larger Study**

The focus of the larger Sleep and Autism study was on investigating the effectiveness of Functional Behaviour Assessment (FBA) informed interventions for sleep problems in children with ASD. Data analysed in the present study were a subset of data taken from the overall larger study.

**The sleep research team.** The larger research project was led by Associate Professor Karyn France, and Dr Laurie McLay, two senior academics at the University of Canterbury, New Zealand. The research team also included PhD candidates, Masters thesis students, Child and Family Psychology interns and registered psychologists.

**Ethical issues.** Ethical approval (reference: HEC 2018/47) was obtained from the University of Canterbury Human Ethics Committee for the larger Sleep and Autism study. No additional approval or consent was required to analyse data included in the present study.

**Participant consent.** Prior to the family's participation in the larger study, all parents were required to give consent for their child's involvement. A brief verbal overview was provided for each family to explain the purpose of the larger study, and what was involved for participants. Each child and family was then provided with written information sheets that included further details. Written consent was received from the parents before the assessment and data collection commenced. Children were verbally informed of the study and what it involved. A child consent or assent form was signed by the parents, on behalf of each child or if children were cognitively and physically able, they signed the consent or assent forms independently. The parent information sheets are shown in Appendix A and the parent consent form is shown in



Appendix B. The information sheet for the children is displayed in Appendix C, the child consent form is shown in Appendix D, and the child assent form is displayed in Appendix E.

**Participants in the larger study.** Participants in the larger study were recruited from organisations throughout New Zealand that provide services for children with ASD, through the networks of the research team, and also by parent self-referral. In total, 54 children and their families participated in the larger study.

Regarding the inclusion/exclusion criteria of participants, the children were eligible for inclusion in the larger study if they: (1) were between 2 and 18 years of age; (2) had been clinically diagnosed with ASD or had demonstrated features of ASD as verified by clinicians; and (3) had parent-reported sleep difficulties (e.g. frequent night waking, co-sleeping, sleep onset delay, or bedtime resistance). Children were excluded from the study if they displayed an existing medical or physical condition that made it unsafe for them to participate in the study (e.g., brain seizure) or that contributed toward their sleep disturbance (e.g., sleep apnoea).

**Materials and measures used in the larger study.** In the larger study, the parent measures used included the DASS-21 that measured mood states, the PSQI that assessed sleep quality, the RQI that measured marital satisfaction, and the Treatment Acceptability Rating Form-Revised (TARF-R; Reimers, Wacker, Cooper, & De Raad, 1992) that estimated the parental perception of the relevance of the treatment. The child measures used were the Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000) that measured sleep problems, the Sleep Assessment Treatment Tool (SATT; Jin, Hanley, & Beaulieu, 2013) that identified the factors that sustained the sleep problems of the child, the Child Behaviour Checklist (CBCL; Achenbach & Rescorla, 2000) that assessed behavioural functioning, the Gilliam

Autism Rating Scale, Third Edition (GARS-3; Gilliam, 2014) that estimated level of ASD severity and the Vineland Adaptive Behaviour Scales-II, Parent/Caregiver Rating Form (VABS-II; Sparrow, Cicchetti, & Balla, 2005) that measured everyday living skills.

**Procedures/study phases in the larger study.** The larger study included six phases: pre-treatment assessment, baseline, intervention phase, post-treatment maintenance, short-term follow-up and long-term follow-up. Baseline typically lasted for 1 to 4 weeks. The intervention phase ranged from 4 to 8 weeks, and this was followed by a post-treatment maintenance period of 4 to 6 weeks. Short-term and long-term follow-up lasted for one week each. A summary of the study phases, measures used and information collected is presented in Table 2.

In the larger study, a semistructured interview was conducted with the parents during the assessment phase. The clinical interview adhered to the format of a standard intake interview as used at the Pukemanu Dovedale Clinic and was administered by an intern psychologist or a researcher who was under the supervision of a registered psychologist. Confidentiality was explained, and informed consent was obtained. The interview focused on gathering information about family characteristics (e.g. sociodemographic information) and parental concerns about the sleep problems of their children. The history and nature of the sleep problems were recorded, and the possibility of conducting an intervention was discussed with the parents. For the purpose of the present study, information about family demographics (e.g. SES) and other family characteristics was obtained retrospectively by examining the clinical interview notes summarised by the psychologists during the semistructured interviews in the larger study. Examples of the clinical interview questions are attached in Appendix F.

***Pre-treatment Assessment.*** The child and parent measures were first administered at pre-treatment assessment to collect pre-treatment information about the sleep and functioning of the children and their parents in the larger study. This information was gathered to inform the treatment plan in the larger study. Parents were prompted to keep their normal practice and maintain the usual sleep routines of the child so as to ensure the accuracy of the collected data in reflecting the real situation.

***Baseline.*** Families were randomly assigned a baseline length of either one, two, three or four weeks. During the baseline phase, parents recorded sleep diaries each night and recorded video on at least 30% of nights. Details from the initial clinical interview and baseline data were used to inform the FBA and, subsequently, to develop a treatment plan. Parents were advised to maintain their usual sleep routines and behaviour during baseline so that any changes in behaviour following treatment could be attributed to intervention strategies rather than to natural variations in behaviour (Blampied, 2013; Kazdin, 1981).

***Intervention.*** The larger study focused on helping the participants with their sleep problems by tailoring sleep interventions to meet the individual needs of each child. The goal of treatment was to decrease sleep interfering behaviours and increase sleep-conducive behaviours. Treatment commenced after the assigned baseline length was due or if a stable pattern on sleep interfering behaviour were detected. Otherwise, the baseline continued until the interfering behaviour appeared stable.

The outcomes of FBA were used to guide treatment planning in the larger study. After integrating the information gathered from all assessments, an initial treatment plan was developed to address the function of the sleep problems for each child as suggested by Blampied (2013). Continuous measures and observations taken

across baseline and treatment phases enabled the therapists to modify the intervention strategies according to the characteristics and changes of a child. A treatment plan based on the outcomes of FBA would address the antecedents or consequences that precipitated or maintained the sleep problems. Interventions included maintenance of sleep hygiene (e.g. teeth brushing), the establishment of positive sleep associations, and manipulation of parental responses (e.g. reduction of parental presence). Other methods also included the use of social stories, stimulus substitution (e.g. replace co-sleeping by sitting next to the child during initial sleep), faded bedtime, camping out, graduated extinction, regular sleep/wake times and weighted blankets.

Daily communication by means of e-mail, telephone, Skype, or text was given to parents for enhancing their motivation and confidence in implementing the new strategies. It also allowed the researchers to resolve any difficulties that arose during treatment. Treatment continued until the sleep problem had resolved and/or the parents were happy with the level of progress, or the parents chose to withdraw from the study.

***Post-treatment maintenance.*** The post-treatment maintenance period began immediately upon the conclusion of the intervention. During this stage, the researchers refrained from contacting the parents, and the parents implemented the programme without the assistance of the researchers (Sanders & Burke, 2014). The post-treatment maintenance period allowed the parents and their children to consolidate the newly acquired behaviours in their daily routines (Blampied, 2013).

A semistructured interview was conducted during the post-treatment maintenance phase to gather information on parental opinions about the treatment and their satisfaction with the treatment outcomes. The child and parent psychometrics

were re-administered at this phase. TARF-R was used to assess the treatment acceptability of the treatment as perceived by the parents.

***Follow-up.*** Short-term follow-up data were collected at 4 to 6 weeks post-treatment (i.e., during the post-treatment maintenance period). Long-term follow-up data were collected at 10 to 12 weeks post-treatment. During the two follow-up phases parents were requested to record one week of sleep diaries and video footage. The follow-up phases aimed to monitor the long-term stability of treatment outcomes.

Table 2

*Summary of Study Phases, Measures used and Information collected*

Phase	Schedule	Duration	Larger study	Present study	
			Measures used/ information collected	Data from larger study being used in present study	Role of the data from larger study that used in present study
Clinical interview			Interview summary SATT	Interview summary SATT	Family demographics (e.g., marital status, occupation) For checking the sleep interfering problems (e.g., sleep onset latency, co-sleeping)
Pre-treatment assessment			DASS-21 PSQI RQI CBCL,CSHQ,GARS-3,VABS-II	DASS-21 PSQI RQI n/a	DASS-D, DASS-A & DASS-S pre-treatment PSQI pre-treatment RQI pre-treatment
Baseline		Between 1 to 4 weeks	Sleep diary Video footage	Sleep diary Video footage	For computation of SPS baseline scores For triangulation of sleep data
Sleep intervention		Between 4 to 8 weeks	Sleep diary Video footage	Sleep diary Video footage	For computation of SPS post-treatment For triangulation of sleep data
Post-treatment maintenance	Upon conclusion of intervention	Between 4 to 6 weeks	Re-administered DASS-21 PSQI RQI CBCL,CSHQ,GARS-3,VABS-II	Re-administered DASS-21 PSQI RQI n/a	DASS-D, DASS-A & DASS-S post-treatment PSQI post-treatment RQI post-treatment
Short-term Follow-up	4 -6 weeks post-treatment	1 week	Post-treatment interview TARF-R Sleep diary	n/a TARF-R Sleep diary	For assessment of treatment acceptability For computation of SPS short-term follow-up score
Long-term Follow-up	10-12 weeks post-treatment	1 week	Sleep diary	Sleep diary	For computation of SPS long-term follow-up scores

**Research design in the larger study.** Treatment outcomes for the children whose sleep problems had been treated were assessed by using a non-concurrent single-case multiple-baseline across subjects design. The larger study within which the children were treated was conducted between 2013 and 2018; in all, a total of 54 children were treated in this period.

***Single-case design.*** Single-case experimental designs are frequently used in behaviour analytic research (Shadish & Sullivan, 2011; Tate et al., 2016) and are appropriate for use in the larger study because such designs permitted the variety of characteristics associated with this heterogeneous group to be retained and analysed. The families in the larger study did not have to wait in a control group, which might reduce the parental stress during the waiting period with no treatment provided (Blampied, 2013; Spruyt & Curfs, 2015).

***Non-concurrent multiple-baseline across subjects design.*** The multiple-baseline across subjects design is a research method commonly used in clinical settings, particularly in cases where multiple clients demonstrate similar behavioural problems (Gast, Lloyd, & Ledford, 2018) in a similar environment (e.g., home environment). Such a design was appropriate for the larger study because it allowed the researchers to analyse the changes in the behaviours of several individuals who have similar problems (e.g., co-sleeping; Blampied, 2014). The multiple-baseline across subjects design involves the replication of a baseline-to-treatment phase change delivered in a staggered structure over time. By delivering the intervention non-concurrently across two or more clients at different time points, each transition from baseline to treatment is a chance to identify any treatment effects (Carr, 2005). The staggered nature of the replications could eliminate the alternative explanations for the behavioural change. Each replication of the intervention, therefore, allows one to

draw an inference that the client's behavioural change is due to the effect of the intervention (Neuman & McCormick, 1995). This helps increase the internal validity of a study (Coon & Rapp, 2018). The numerous inter-subject replication characteristic of the multiple-baseline across subjects design also enhances the external validity of a treatment (D.L.Morgan & R.K.Morgan, 2008).

**Data analyses in the larger study.** In the larger study, data obtained using a combination of video footage, sleep diaries across study phases was graphed for visual inspection, according to the target behaviours for each child. The data of the parent-completed sleep diaries were first entered into the Excel spreadsheets and then graphed according to the common sleep problems across children. This included the duration of sleep onset latency, the frequency of curtain calls, the frequency of night waking, and duration of night waking. Visual inspection was used as the primary means of data analysis as a comparison between study phases. Visual inspection of the graphs included analysis of change in direction, variability, length, and consistency (Cohen, Feinstein, Masuda, & Vowles, 2014).

### **The Present Study in the Context of the Larger Study**

The aim of the present study was to investigate whether family complexity components could predict the treatment outcomes of children with ASD. To achieve this, the present analysis also aimed at examining whether there were latent components residing underneath the 11 parent/family variables of family complexity.

**Participants in the present study.** For the present study, participants were selected from the larger study if they had completed: (1) the initial assessment and clinical interview, (2) pre-intervention psychometrics, (3) baseline sleep diaries, and (4) post-treatment psychometrics and sleep diaries. Families that dropped out of the



larger study before the completion of intervention were excluded from the data set of the present study.

Data collected for 31/54 participants from the larger study were eligible to be included in the data set for statistical analysis in the present study. The participants included 7 girls and 24 boys aged between 3 and 14.5 years of age. A summary of participant characteristics is presented in Table 3.

Table 3

*Summary of Participants Characteristics (n = 31)*

Characteristics	Values
Parent and family	
Single-parenthood	36%
Low SES	39%
Parental psychopathology	29%
Parental criminality	3%
Family with 3/+ children	32%
Siblings had ASD or DD	7%
Resided outside the district of the clinic	61%
Child	
Age range (mean)	3 – 14.5 (6.59) years old
Boys	77%
Clinical ASD diagnosis	97%
With more than one type of DD	48.0%
Current on psychotropic medication	39%
Presenting Sleep problems	
Bedtime resistance	32%
Sleep onset delay	65%
Night waking	84%
Early waking	48%
Sleep interfering behaviour	71%
Co-sleeping	55%

**Materials and measures used in the present study.** To make this section more relevant to the research aims of the present study, only those materials and measures that were relevant to the present study, used to assess the parent/family functioning, or utilised to calculate the sleep problem severity of the children are described in this section. The parent measures that included DASS-21, PSQI, RQI and TARF-R are explained in the following paragraphs, followed by a composite family adversity score. The child measures that included a composite sleep problem severity (SPS) score, sleep diaries, video footage and SATT are also presented.

***Parent/family measures.*** The parent/family measures included DASS-21, PSQI, RQI and TARF-R. As mentioned earlier, the outcome scores of DASS-21, PSQI and RQI at pre-treatment were included in the 11 parent/family variables that defined family complexity in the present study. A composite family adversity score was developed to indicate the level of adversity a family was experiencing prior to treatment. This family adversity score was one of the 11 parent/family variables. The TARF-R was used to check the social validity of the sleep intervention provided for the families.

***Depression Anxiety and Stress Scale-21***(S.H. Lovibond & P.F. Lovibond, 1995). The DASS-21 is a short version of the 42-item self-report index of depression, anxiety and stress. The 21 items of DASS-21 assess levels of depression (DASS-D), anxiety (DASS-A) and stress (DASS-S) of an individual. Each dimension has seven statements arranged on a 4-point Likert scale ranging from 0 (*never*) to 3 (*almost always*) to indicate the emotions experienced in the past week. The score on each of the seven items is then added to yield a total score for that scale. By multiplying the total score by 2, a subscore for that scale is produced. The possible score range for each subscale is from 0 to 42. A higher score denotes a higher level of distress. For

each subscale, the level of distress can be subdivided into *normal*, *mild*, *moderate*, *severe* and *extremely severe categories*. Each category has a range of scores. For the depression scale, a subscore of more than 9 denotes mild symptoms of depression. If a subscore of anxiety is higher than 7, it indicates the presence of anxiety. A score of 15 or above on the stress subscale means that the individual is experiencing a mild level of stress (S.H. Lovibond & P.F. Lovibond, 1995). The reliability coefficients of the three subscales have been found to range from .82 to .97, which indicates good internal consistency across DASS-21 items (Henry & Crawford, 2005; P.F. Lovibond & S.H. Lovibond, 1995). The DASS-21 was administered pre- and post-intervention in the larger study. The collected data were used to determine the mood states of the parents, and were referred to as paternal DASS-depression (DASS-D), paternal DASS-anxiety (DASS-A), paternal DASS-stress (DASS-S), maternal DASS-depression, maternal DASS-anxiety and maternal DASS-stress in the 11 parent/family variables in the present study.

*Pittsburgh Sleep Quality Index* (Buysse et al., 1989). The PSQI is a 19-item self-report questionnaire that is used to assess the severity and frequency of symptoms associated with major sleep disorders. These 19 items are grouped into seven subscales; each weighted equally on a 0 (*no difficulty*) to 3 scale (*severe difficulty*) and are summed to yield a total score ranging from 0 to 21. Higher total scores (referred to as global scores in the original PSQI validation study), indicated poorer sleep quality (Buysse et al., 1989). A PSQI total score of more than 5 is indicative of poor sleep (Buysse et al., 1989). A global score that is greater than 5 has 89.6% sensitivity and 86.5% specificity for distinguishing good and poor sleepers (Carpenter & Andrykowski, 1998). The PSQI correlates moderately to very well with other scales of sleep quality and sleep problems (Carpenter & Andrykowski, 1998). In the

larger study, the PSQI was administered during pre- and post-treatment phases. The collected data were used to determine the sleep quality of the parents, and were labelled as paternal PSQI and maternal PSQI in the 11 parent/family variables in the present study.

*Relationship Quality Index* (Norton, 1983). The RQI is a 6-item self-report questionnaire that assesses marital quality and satisfaction. The first five statements are arranged on a 7-point scale from 1 (*very strongly disagree*) to 7 (*very strongly agree*). The sixth statement is measured on a 10-point scale (1 = *unhappy*, 10 = *perfectly happy*). Scores of less than 29 are indicative of relationship distress (Heyman, Sayers & Bellack, 1994). The scale demonstrated high internal consistency in studies recruiting parents of children with ADHD ( $\alpha = .98$ ) (Hoath & Sanders, 2002), toddlers with behaviour problems ( $\alpha = .95$ ) (Morawska & Sanders, 2006) and children with ASD ( $\alpha = .94$ ) (Roux, Sofronoff & Sanders, 2013). The RQI was administered pre- and post-treatment in the larger study. The collected data were used to estimate the relationship quality of the parents, and were referred to as the paternal RQI and maternal RQI in the 11 parent/family variables in the present study.

*Treatment Acceptability Rating Form-Revised* (Reimers et al., 1992). The TARF-R is a 20-item questionnaire used to provide a global rating of treatment acceptability (Reimer et al., 1992). The TARF-R measures treatment acceptability across eight dimensions. This is then used to derive a total acceptability score which is the sum of the scores of 17 items that measure the six dimensions of acceptability (i.e. reasonableness, effectiveness, side effects, disruptive/ time consuming, cost, and willingness). Two additional items address problem severity, and one item measures the respondent's understanding of the intervention. The items are arranged on a 7-point Likert-scale from 1 (*not at all*) to 7 (*very much*). Reimers et al. (1992) classified

the total acceptability score into *high acceptability* (scores ranging from 85 to 119), *middle acceptability* (scores ranging from 52 to 84), and *low acceptability* (scores ranging from 17 to 51). The TARF-R has high internal consistency ( $\alpha = .92$ ). The parent's acceptability of the intervention was administered at the post-treatment phase in the larger study. For the purpose of the present study, the TARF-R scores were correlated with treatment outcomes for each child in order to determine whether the parental perception of treatment acceptability was related to children's treatment outcomes. The labels paternal TARF-R and maternal TARF-R were used.

*Family adversity score.* In the present study, family adversity was included in the 11 variables that defined family complexity. The family adversity score was a composite score that addressed the intensity of adversity a family was experiencing at the pre-treatment phase. The items of the family adversity score were based upon that used by Rutter et al., (1975) and Rutter and Quinton (1977). The adversity variables included: (1) *marital conflict*, (2) *family SES*, (3) *family size*, (4) *parental psychopathology*, (5) *parental criminality*, (6) *siblings of the participant had developmental disabilities*, (7) *geographical barriers*, (8) *disagreement between parents about the treatment*, (9) *parent-therapist communication difficulties*, and (10) *other barriers to treatment participation*. Data on the first seven of these adversity items were collected during the clinical interview process. Data on the final three was also gathered during the treatment process. Clinical interview notes were examined, and a conference discussion with the psychologists involved in the cases was held to discuss whether a family displayed incidence of these ten adversity items.

To calculate a total adversity score, a rating of 1 or 0 was assigned to each variable to denote the presence or absence of an adversity item (Rutter et al., 1975; Rutter & Quinton, 1977). Therefore, the total overall family adversity score ranged

from 0 to 10. A higher score represented a higher level of adversity. A family adversity score was calculated for conditions existed at pre-treatment or before intervention conclusion. No post-treatment adversity score was calculated as this was not necessary to determine any interaction between adversity and treatment outcomes. A description of each of the family adversity variables is provided below:

(1) *Marital conflicts*: Marital conflicts were determined by the presence of parental conflict or domestic violence as reported by the parents in the clinical interview or revealed by the psychologists in the conference discussion. Marital conflict was defined as any interaction between the two parents in response to a difference of opinion involving destructive effects on the relationship and strong negative emotionality (Cummings, Goeke-Morey, & Papp, 2004). Domestic violence was defined as any form of physical or verbal violence experienced by the individual (Richardson et al., 2002). Presence of any form of reported parental conflict or domestic violence was scored as 1. A score of 0 denoted no parent-reported incidence of marital conflict.

(2) *Socioeconomic status*: The New Zealand Socioeconomic Index 2013 (NZSEI-13) was used to determine the SES of each of the families (Fahy, Lee, & Milne, 2017). The NZSEI-13 is the latest version of the New Zealand socioeconomic index, and is based upon the 2013 Census data. The NZSEI-13 score, which is an occupation-based measure of SES, was constructed on data from both full-time and part-time employees, with income adjusted for those engaged in part-time jobs. The precursor of NZSEI was the widely-used Elley-Irving scale (Elley & Irving 1972; 1976; 1985; 2003; Irving & Elley, 1977), that allocated occupations to one of the six socioeconomic categories based on equal weighting of the education level and salary associated with each occupation. An assessment was undertaken to explore whether

the NZSEI-13 methodology allocated scores similarly for men and women, different ethnicities, urban and rural workers, various geographical regions, and different countries of birth. The results showed that, although there were some differences in average scores, occupations were categorised very similarly by sex-specific, ethnic-specific, region-specific, and country-of-birth-specific scales. This suggests that NZSEI-13 applies to both sexes, various ethnic groups (e.g. New Zealand European, Maori, and Asian), urban and rural workers, workers in Auckland and the rest of New Zealand, and both workers born in New Zealand and workers born overseas. A comparison between NZSEI-13 and the earlier NZSEI-06 showed that both scales classified individuals from the 2013 New Zealand Census with the almost identical result (correlation:  $r > .99$ ; Fahy et al., 2017). The NZSEI-13 score ranges from 10 to 90, with packers and product assemblers scoring the lowest (i.e., 10) and medical practitioners scoring the highest (i.e., 90). In the present study, the operational definition of low social class would be a NZSEI-13 score between 10 and 39. On this basis, a family having a NZSEI-13 score of 39 or below was scored 1 in the family adversity composite score. A family having a NZSEI-13 score of 40 or above was scored 0.

(3) *Family size*: Large family size was defined as three or more children in a family (Mick, Biederman, Faraone, Sayer, & Kleinman, 2002). Only those siblings who regularly lived with the child with ASD and their mother were included in family size estimation. A family was scored 1 if there were three or more children in a household. A score of 0 was assigned if the family had one or two children.

(4) *Parental psychopathology*: Parental psychopathology was deemed present if the mother or the father had at least one psychological disturbance (e.g. depression or substance dependence) during the child's lifetime, including the prenatal period



(Mick et al., 2002). The psychological disturbance included diagnosed disorders or parent self-reported symptoms. For instance, a score of 1 was assigned if there was an incidence of parent-reported psychopathology. Zero was assigned when no incidence of psychopathology was reported.

(5) *Parental criminality*: An incidence of jail confinement in either the father or the mother was assigned a score of 1 to the family adversity score to index parental criminality. A score of 0 was assigned when no incidence was reported.

(6) *Sibling(s) with developmental disabilities*: A family had more than one child with diagnosed developmental disorders or developmental delay was scored 1. The disabilities also included symptoms of ASD, pervasive developmental disorders and other development disabilities. Zero was assigned for no sibling had these problems. Since the siblings of the participants had no report of having physical disability, physical disability was not considered in this category.

(7) *Geographical barriers*: The larger study was based in the Pukemanu Dovedale Centre Clinic at the University of Canterbury, Christchurch, New Zealand. The present study assumed that families living in other regions of New Zealand may find it more difficult to access the clinic and get the same level of support from the sleep team when compared to those families living within the Canterbury region. Kazdin, Holland, Crowley and Breton (1997) also suggested that living out of the clinic area could be a barrier to treatment participation for some clients. Because of this, a family was assigned a score of 1 if the residential address was outside Canterbury to denote the presence of geographical barriers whereas a score of 0 was allocated to those families residing in Canterbury.

(8) *Disagreement between parents about the treatment*: Disagreement between

the two parents was defined as a lack of consensus in substantial and procedural matters of the intervention (Teven, McCroskey, & Richmond, 1998). A score of 1 was assigned to the family if there were any disagreements between the two parents regarding the best approach to treatment. The scoring decision was based upon reports from the psychologists working with the parents. A score of 0 was assigned for no presence of parental disagreement.

(9) *Parent-therapist communication difficulties*: Parental behaviours such as being repeatedly late for appointments, being difficult to engage, and hard to reach have been regarded as communication barriers to behavioural programmes for childhood behaviour problems (Doherty, Stott, & Kinder, 2004; Forehand et al. 1983; Kazdin Holland, & Crowley, 1997; Kazdin Holland, Crowley, & Breton 1997; Koerting et al., 2013). Kazdin, Holland, Crowley, & Breton (1997) have suggested that parent-therapist communication barriers might include a client does not like the therapist, a client does not have confidence in the treatment or the therapist, a client does not feel supported by the therapist, and a client feels that the therapist does not call often enough. In the present study, barriers related to the communication with the therapists included (1) being repeatedly late for appointments, (2) being difficult to engage, and (3) being hard to reach. In addition, in occasional cases, client would report not liking the therapist, not having confidence in the treatment, not feeling sufficiently supported by the therapist. Parent-therapist communication barrier was regarded as present in a family if the psychologists revealed the aforementioned parental behaviours or statements in the conference discussion. A score of 1 was assigned for the presence of parent-therapist communication barriers whereas a 0 for an absence of features.

(10) *Other barriers to treatment participation*: Issues with treatment

attendance or treatment adherence may affect the treatment outcomes (Forehand et al., 1983; Kazdin, Holland, Crowley et al., 1997). Treatment attendance was defined as the presence of treatment participants (e.g., parent, child, family, etc.) to the treatment setting (e.g., at clinic, at home or in the telephone session) for scheduled appointments (Nock & Ferriter, 2005). In the context of the present sleep study, parental absence in scheduled appointments at the clinic, at home or telephone/skype sessions was regarded as the presence of difficulties in treatment attendance. Parental treatment adherence was defined as active, voluntary, collaborative parental involvement in a mutually agreed course of behaviour or intervention strategies designed to produce a desired therapeutic result (Meichenbaum & Turk, 1987). In the present study, barriers to treatment adherence could include parental reluctance to change, parents had an inappropriate attribution about the child's sleep problem, parental non-compliance to treatment plan or parental difficulty in completing sleep diaries. Any reported incidence of the above mentioned parental behaviours was treated as the presence of a barrier in the family, and a score of 1 was allocated. A score of 0 was assigned for the absence of these behaviours.

The sum of the above ten subscores would produce an *overall family adversity score*. The work of Rutter et al. (1975), and Rutter and Quinton (1977) have suggested that it might be the combination of multiple adversity variables, instead of the presence of any single family factor that impairs the development of a child. The findings of the work of Rutter et al. (1975) and Rutter and Quinton (1977) indicated that no single family factor significantly predicted the risk of developing psychiatric disorders or social deviance, but the presence of two family factors resulted in a fourfold increase in the risk. The presence of four family factors yielded a tenfold increase in the risk of developing psychiatric problems and social deviance

(Biederman et al. 1995). Similar to the method used by Rutter et al. (1975), and Rutter and Quinton (1977), in the present study, the operational definition of the family adversity was the summation of the 10 family adversity subscores that permit a single metric (i.e. the overall family adversity score) (Tarling & Perry, 1985).

**Child sleep measures.** For the purpose of the present study, the data of sleep diaries and video-recording were used for calculation of SPS. The SATT was used to check the sleep disturbance the child experienced when they participated in the larger study.

*Sleep problem severity score* (Lawton, France, & Blampied, 1991). The SPS score was a composite score developed to operationally define the severity of sleep problems in the participants. The SPS score was calculated for each child at each study phase. Therefore, each child would have four SPS scores if they had completed all four phases. They are SPS baseline (SPS BL), SPS post-treatment (SPS Post-tx), SPS short-term follow-up (SPS STFU) and SPS long-term follow-up (SPS LTFU). To quantify the treatment outcome, the change in SPS scores between two time points was computed. By that, each child would have three SPS change scores if the child had completed all four phases. They are, SPS baseline vs SPS post-treatment (SPS BL vs SPS Post-tx), SPS baseline vs SPS short-term follow-up (SPS BL vs SPS STFU) and SPS baseline vs SPS long-term follow-up (SPS BL vs SPS LTFU).

The SPS scores were calculated using the final seven nights of baseline, the final seven nights of treatment, and the seven nights of short-term follow-up and the seven nights of long-term follow-up. The SPS calculation system was based on the mechanism used by Lawton et al., (1991) and Richman (1985) for assessing the sleep behaviour of infants. SPS scores were also adapted to reflect the developmental norms of school-age children and adolescents. As a result, three sets of scoring system were

formulated according to three different age groups; they are pre-schoolers (ages 3 to 4 years 11 months), primary school-age children (ages 5 to 12 years) and adolescents (ages 13 years and over). The scale was also modified to accommodate the nature of the present study. For instance, parental presence and sleep setting were added to address the sleep interfering behaviours of the children. The SPS scoring guides for each age group are shown in Appendix G (pre-schoolers), Appendix H (school-age children) and Appendix I (adolescents).

The SPS score was comprised of nine elements, as follows: (1) *bedtime each night*, (2) *time taken to fall asleep each night*, (3) *total time slept each night in hours*, (4) *number of wakings each night*, (5) *average time awake per waking (minutes) each night*, (6) *percentage of night spent co-sleeping each night*, (7) *parental presence for initial sleep onset*, (8) *parental presence during sleep onset following night waking*, and (9) *sleep setting*. Regarding the scoring of the first six elements, a child received a score ranging from 0 to 4 depending on the degree of occurrence. The higher the score, the more severe or frequent the feature. For the last three elements, the scores were dichotomised into 0 (absence of the feature) or 1 (presence of the feature). The sleep diary and video footage were the main sources of data used to calculate the subscores of SPS. The scoring of each element of SPS are described as follows:

(1) *Bedtime each night*: The time of *bedtime each night* was defined as the time that once the child was put to bed (Wolke, Meyer, Ohrt, & Riegel, 1995). In the sleep diary, it was named as the *time put to bed*, which was felt more easily understood by the parents. For instance, for a school-age child (5 to 12 years old), bedtime at 9:29 p.m. or earlier was scored 0 whereas bedtime after 9:30 p.m. was scored 1.

*Best estimate of asleep time:* In the sleep diary, the *best estimate of asleep time* indicated the time (e.g., 7:15 p.m.) that the child fell asleep recorded by the parents. Indicators of asleep included eyes closed, minimal movements and absence of voluntary vocalisation (Carskadon, 1986). The information of this item was used together with *time put to bed* to estimate the *time taken to fall asleep*.

(2) *Time taken to fall asleep each night:* The *time taken to fall asleep each night* was regarded as the duration of sleep onset latency. Sleep onset latency was defined as the time duration that a child took to achieve the transition from full wakefulness to initial level of sleep (Weiner & Craighead, 2010). In the sleep diaries, the parent marked the *time put to bed* and *best estimate of time asleep* of the child. The sleep onset latency was the difference (in minutes) between these two time points. For example, if *time put to bed* was 7 p.m. and *best estimate of time asleep* was 7:30 p.m., the *time taken to fall asleep* was 30 minutes. For a school-age child, a less-than-15 minutes duration of sleep onset latency was scored 0. However, a duration of more than 60 minutes was scored 4.

With regard to *time taken to fall asleep each night* and *bedtime each night*, either one was included in the total SPS score depending on whichever was worse. For example, a pre-schooler (3 to 4 years 11months) who took 40 minutes to fall asleep was scored 2 while ‘bedtime at 7:30 p.m.’ was scored 0. In such a case, the higher score was selected to reflect the more serious condition; thus, *time taken to fall asleep each night* (i.e., 2) was included in the calculation of the total SPS score.

When the treatment plan advised parents to delay their child’s bedtime (i.e., bedtime fading), the *time taken to fall asleep each night* would be used instead of *bedtime each night*. Bedtime fading, a strategy of delaying a child’s sleep time, would increase the child’s need to sleep, and would eventually reduce the child’s reluctance

to sleep (France & Blampied, 2005). The *bedtime each night* would be manipulated so as to purposefully alter the child's bedtime. In this case, the altered bedtime was an artificial bedtime which did not mean a worse condition. Therefore, the *time taken to fall asleep each night* would be included in the calculation of the total SPS score.

(3) *Total time slept each night*: The *total time slept each night in hours* represented the total sleep hours a child had over a night. The parent recorded the child's *time awake in the morning*. *Time awake in the morning* represented the time that the child woke up in the morning with readiness for daily activities (e.g., going to school). Awake was defined as arousal with eyes open, vocalisation of any sort, or excessive physical movement in the bed or under the covers (Jin et al., 2013). The total sleep time was the difference (in hours) between *best estimated time asleep* and *time awake in the morning*.

The sleep duration was categorised in accordance with the age-specific recommendation stated in the National Sleep Foundation Guidelines (Richman, 1985). For instance, a pre-schooler who slept more than 10 hours would gain a subscore of 0 while sleeping less than seven hours would yield a score of 4. In contrast, a primary school-age child, sleeping more than nine hours would gain a subscore of 0 while sleeping less than six hours would produce a subscore of 4 (Lawton et al., 1991).

(4) *Number of wakings*: The *number of wakings each night* indicated the frequency of night waking throughout a night. The parent logged the number of times the child demonstrated night waking in a night. A night waking was defined as an episode of arousal in which the child did not self-reinitiate to sleep (Henderson et al., 2010; Knight & Johnson, 2014). For a child, a night without night waking was scored 0 whereas a night with 4 or more times of night waking was scored 4.

(5) *Average duration of night waking*: The *average time awake per waking (minutes) each night* denoted the average length (in minutes) of each night waking in a night. For instance, if a child had three times of night waking in a night, with one lasting for 4 minutes and two for 10 minutes each, the total duration of night waking would be 24 minutes of that night, with an average length of these three episodes of night waking was 8 minutes. For a school-age child, an episode of night waking with an average duration of more than 60 minutes was scored 4.

(6) *Percentage of night spent co-sleeping*: The *percentage of night spent co-sleeping each night* was calculated based on the portion of the time the parent spent co-sleeping with the child in a night. Co-sleeping was defined as the child sleeping in the same bed with an adult within a close distance that allowed the exchange of any two types of sensory stimuli such as touch, movement, smell or sound for any period of a night (McKenna & Volpe, 2007). For instance, if the parent slept two hours co-sleeping with the child, and the child had eight hours of sleep on that night, the percentage of co-sleeping was 25%. For a school-age child, 1 to 20% of co-sleeping each night was scored 1 whereas co-sleeping of more than 61% was scored 4.

(7) *Parental presence during initial sleep onset*: The *parental presence during initial sleep onset* denoted whether the parent stayed with the child at bedtime until the child fell asleep (Wolke et al., 1995). A subscore of 1 was added to the total SPS score if the parent was either in physical or visual proximity to the child.

(8) *Parental presence during sleep onset following a night waking*: Similar to the parental presence at sleep onset, if the parent stayed in physical or visual proximity to the child following a night waking (Wolke et al., 1995), a subscore of 1 was added to the total SPS score.



(9) *Sleep setting*: The *sleep setting* item denoted whether the child slept in his/her own bed or not. For instance, if the child slept on the couch or the bed of the parents, a subscore of 1 was assigned. A 0 score was assigned when the child slept in his/her own bed.

To calculate the total SPS score of a single night, the sum of 9 subscores of one night would yield a total SPS score of that night. To calculate the final SPS score for each child, in one study phase, the calculation of the total SPS score of a single night was replicated in seven consecutive nights of the phase. Hence, there would be a total of seven SPS scores. The sum of these seven scores was then divided by seven to produce an average SPS score. This average SPS score was treated as the final SPS of a study phase. These steps were repeated in each phase, and therefore, in total, four SPS final scores would be available (i.e., SPS baseline, SPS post-treatment, SPS short-term follow-up, and SPS long-term follow-up). The possible range of a SPS score would be from 0 to 23. A higher score indicated a more severe sleep problem.

The treatment outcome was operationalised as the change in SPS scores between baseline and the other three time points. The three SPS change scores were (1) SPS baseline vs SPS post-treatment score which denoted the treatment outcome at post-treatment; (2) SPS baseline vs SPS short-term follow-up score which indicated the treatment outcome at short-term follow-up; and (3) SPS baseline vs SPS long-term follow-up score which represented the treatment outcome at long-term follow-up. The three SPS change scores were treated as dependent variables to be predicted by parent/family predictors.

A greater value of SPS change score would denote a bigger improvement in sleep problems. A negative SPS change score would imply that the level of sleep problem severity had worsened. For instance, if a child had a SPS baseline score

higher than the SPS short-term follow-up score, it would mean that the child demonstrated a reduction in sleep problem severity. Likewise, if the SPS score increased between baseline and post-treatment, it would represent an increase in sleep problem severity. The SPS score was the sum of subscores of the aforementioned nine items such as total sleep time, the percentage of co-sleeping, etc. On this basis, improvement in sleep problem could mean increased total sleep time, shortened sleep onset latency, reduced percentage of co-sleeping or diminished parental presence.

The SPS scores were calculated by the researcher and another master thesis student, and compared for reliability. In general, the data of the final seven consecutive nights of each phase were used. When data of some items of one night were found missing in the sleep diaries, the video footage of that particular night would be re-addressed and missing information was re-coded. If the video footage were also found unclear or missing, the latest available night right before the final seven nights would be used to replace the night with missing data. For the two follow-up phases, there were only seven nights in each phase. If data of one night were not available, the average of the data of the other nights of the week would be used to substitute the missing data of that unavailable night.

*Sleep diaries.* Sleep diaries were provided to the parents for recording information about (1) daytime sleep, including setting, time asleep and time awake; (2) sleep routines (time put to bed, time awake to commence the day); (3) night-time sleep (setting, time put to bed, the frequency, nature, parental response to curtain calls and time until silence); (4) night waking (time, duration, and frequency of awakening, the child's behaviours while awake, and parental responses); and (5) the time that the child wakes up in the morning. The sleep diaries were formatted so that the parents could either enter data by circling a code or writing a description of behaviours that

occurred. Behaviour codes were used to record the child's behaviours and parental responses that commonly occurred for children with sleep problems. This enabled the parents to record the behaviours quickly. For example, the code *L* was used to denote the parent laid down with the child, and *C* represented that the child cried. A copy of a typical sleep diary is attached in Appendix J.

In the larger study, during the baseline phase, diaries were collected at least once a week. During the intervention phase, the researcher made daily contact with the families to collect sleep diaries so that progress could be closely monitored. During follow-up, sleep diaries were collected on completion of the week of recording. Upon receiving sleep diaries, data were entered into an Excel spreadsheet and visually analysed. Sleep diaries have been used frequently in previous research to obtain data on children's sleep (Blampied, 2013; France & Blampied, 2005; McLay, France, Knight, Blampied & Hastie, 2019). In the present study, the information recorded in sleep diaries was the primary source of data for computing the SPS scores.

*Video recordings.* Video recordings are commonly used in sleep studies to assess the sleep profile of children (Blampied, 2013; Hanley, Jin, Vanselow, & Hanratty, 2014). In the larger study, video recordings were collected for a minimum of 30% of nights across study phases. The video recordings helped triangulate data collected through sleep diaries and sleep psychometrics. This was a mechanism for minimising parental bias and quantifying sleep-interfering behaviours (Knight & Johnson, 2014; Spruyt & Curfs, 2015). Video recordings were used to code information including sleep onset time, co-sleeping frequency, frequency and duration of night waking, activities of parents or child while awake, and wake up time. The data of recordings were also used for the calculation of inter-observer agreement. In the present study, when a piece of data recorded by the parents in sleep diaries was

found to be ambiguous or insufficient, relevant video footage was coded to permit the cross-checking of data and/or fill in any missing information. The video recording consent form is shown in Appendix K.

*The Sleep Assessment Treatment Tool* (Jin et al., 2013). The SATT was administered during the initial clinical interview to guide the FBA process. The SATT has been repeatedly used to inform the development of sleep treatment plans in children with ASD (Jin et al., 2013; McLay, France, Blampied, Danna, & Hunter, 2017). The SATT is an open-ended interview tool designed for identifying the sleep problems and the idiosyncratic environmental factors contributing to the child's sleep disturbance (Hanley, 2015). Specifically, the SATT gathers information about (1) the types of sleep problems (e.g., sleep onset delay), (2) antecedent and consequence factors that precipitate or maintain the sleep problem, (3) the history of the child's sleep problems, (4) parents goals for treatment, (5) the child's sleep routine and schedule, (6) the sleep environment, and (7) the type of sleep interfering behaviours and sleep dependencies. The information collected was used to help construct an individualised intervention that was appropriate for each child and family. For the purpose of the present study, the SATT data were used to examine the nature and number of sleep disturbance that a child possessed.

**Research design in the present study.** The present study is a follow-up subset data analysis of the larger study; therefore, basically, the research design is the same as that in the larger study. That is, a combination of single-case design and non-concurrent multiple-baseline across subject design. Details have been explained in the earlier section that described the research design of the larger study, and therefore, are not repeated in this section.

**Data analysis in the present study.** The scores of child sleep problem severity (i.e., SPS) and parental mood states (i.e., DASS-21), parental sleep (i.e., PSQI), parental relationship (i.e., RQI), and family adversity were all inputted into an Excel spreadsheet as well as Statistical Package for the Social Sciences (SPSS) spreadsheets to permit analysis of the relationship among these variables. All data were analysed by using the SPSS 25.0 for Windows 10 (SPSS Inc., Chicago, 2017).

For the purpose of the present study, the pre- and- post comparison method was used to examine whether the behavioural sleep treatment was effective in reducing the sleep problem severity of the children by comparing the SPS scores obtained at baseline against another time point following treatment (i.e., Research Question 1). The pre- and post- comparisons on parental measures were also computed to investigate any secondary effects of the treatment.

In order to explore the underlying components that lay underneath the 11 parent/family variables of family complexity, the 11 parent/family variables obtained at pre-treatment were firstly analysed by using factor analysis, and then the extracted factors (i.e., the three components of family complexity) identified were examined via a correlational approach (i.e., Research Question 2). After that, multiple regression analysis was used to determine if the family complexity components could predict the treatment outcomes (i.e., Research Question 3).

**Modified Brinley plots.** Our Research Question 1 focused on investigating whether the behavioural intervention was effective in reducing the sleep problem severity in the children. Modified Brinley plots were used to determine whether children's sleep problem severity reduced following the intervention (Blampied, 2017). Modified Brinley plots displayed individual changes in SPS scores at two time-points, for example, between baseline and post-treatment, or between baseline

and short-term follow-up. The advantage of Modified Brinley plots is that the performance of each individual can be clearly presented within one figure (Blampied, 2017). Modified Brinley plots were used as tools for the visual inspection of therapeutic changes.

***Wilcoxon signed-rank test.*** The Wilcoxon signed-rank testing was used for the statistical computation of changes between two time-points. Taking the ordinal nature of the composite SPS score into account, the Wilcoxon signed-rank test was an appropriate non-parametric test to be used in the present study (Ho, 2006).

***T-test.*** With regard to the secondary outcomes, we investigated whether parental mood states, sleep quality and marital relationship would improve following intervention. We examined the scores of DASS-21, PSQI and RQI obtained at pre-treatment and compared to those obtained at post-treatment. The *t*-test was used for the statistical computation of the changes between two time-points.

***Spearman's rank order correlation.*** The parametric assumptions were not entirely fulfilled because of the nature of some variables. For example, SPS scores were scored based on ordinal level scaling and because of this, Spearman's rank order correlation, a non-parametric test, was used for correlation computations. To understand the overall picture of the relationship among the child and parent/family variables before further investigating the research questions 1, 2 and 3, we first examined whether different parent psychometrics correlated with one another (e.g., between DASS-21 and RQI), and whether parent psychometrics were related to child sleep problem severity (i.e., SPS scores). It was anticipated that some correlations would be negative and some would be positive. For instance, poor parental sleep quality (i.e., a higher score of PSQI), may be positively correlated with children's sleep problem severity (i.e., a higher score of SPS). Likewise, ratings of relationship

satisfaction (i.e., high RQI score), may be negatively correlated with ratings of mood states (i.e., low DASS-21 subscores).

Spearman's rank order correlation also enabled the investigation of whether parental perceptions of treatment acceptability were related to treatment outcomes of the children (i.e., Research Question 4). The present study investigated the relationship between treatment outcomes (e.g., the change in SPS between baseline and post-treatment) and ratings of treatment acceptability. The correlations between the SPS change scores and the total score of TARF-R were analysed to identify any relationship between treatment outcomes and treatment acceptability. It was anticipated that a greater reduction in sleep problem severity (i.e., a greater SPS change score), would be associated with high social validity ratings (i.e., a higher score of TARF-R).

***Exploratory factor analysis.*** Research Question 2 focused on identifying the components of family complexity that underlie the ten parent variables and one family adversity variable collected at pre-treatment assessment phase. Exploratory factor analysis (EFA) was used to determine the components resided underneath the 11 parent/family variables of family complexity, and this allowed the uncovering of the underlying structure of a relatively large set of variables, that is, the 11 parent/family variables in the present study. The method of EFA enabled us to extract maximum variance from the data set with each component involving a cluster of extracted variables. The EFA computation would result in reducing a large number of variables to a smaller number of components or domains (Tabachnick & Fidell, 2007). From this, the minimum number of components to be retained in family complexity that accounts for maximum variance in the data could be retained. The retained components were used for subsequent multivariate analysis.

In the present study, the use of principal components analysis (PCA) produced eigenvalues that represented the variance accounted for by each underlying component. The eigenvalues enabled the selection of which and how many components should be retained for the computation of factor scores. The PCA extraction method also produced factor loadings for every extracted variable (e.g., maternal PSQI) within each of the family complexity components (Tabachnick & Fidell, 2007). The factor loadings of the clusters of extracted variables within a component were used for the computation of a factor score for each of the retained components. Varimax rotation method was used in the present study because it increased the variances of the factor loadings and thus produced both the large and small factor loadings. With that, the variables were clearly loaded or not load onto each factor, which permitted a clear structure of underlying components to be obtained (Howard, 2016). After the extraction of the latent components of family complexity, a factor score was computed by using the factor loadings of all variables that belonged to a component. For example, if three components were extracted, each child was assigned with three factor scores.

***Multiple regression analysis.*** In order to investigate whether components of family complexity could predict treatment outcomes in the children (Research Question 3), multiple regression analysis was used for examining the prediction of treatment outcomes by multiple family complexity components. A multiple regression analysis was appropriate for this analysis because, in each computation of prediction, multiple independent variables (also known as predictor variables) and a single dependent variable (also known as criterion variable; Dancy & Reidy, 2004) were involved. The advantage of multiple regression analysis is that it allows for a more precise understanding of the association of each individual family complexity



component with the treatment outcomes (Marill, 2004). A simultaneous multiple regression analysis was used because it was anticipated that several family complexity components would be extracted from the factor analysis. Based on the simultaneous model of analysis, all of the independent variables (i.e., factors scores converted from each family complexity component) were entered simultaneously to determine whether independent variables could predict treatment outcomes (i.e., the SPS change scores). There was no theoretical basis or empirical evidence from current studies for deciding any extracted component to be prior to any other, and because of this, the simultaneous regression method was suitable in the present analysis.

## **Results**

### **Chapter 4**

This chapter presents the data analyses that investigated the treatment effects of the home-based sleep behavioural intervention, and the relationships between components of family complexity and treatment outcomes of 31 children with ASD. The section begins with an overview of frequency distributions of the child SPS scores followed by the frequency distributions of three measures of parent functioning (i.e., DASS-21, PSQI and RQI). The descriptive statistics of the child SPS scores at four time points as well as the three SPS change scores are then illustrated. Treatment effects are also interpreted by using modified Brinley plots and the Wilcoxon signed-rank test. Descriptive statistics and *t*-test comparisons of pre- and post-treatment scores of the three parent measures are then presented. The results of Spearman's rank correlations between parent/family variables and child SPS scores are also reported. The three components of family complexity extracted by using exploratory factor analysis are described, The results of multiple regression analysis using to examine the prediction of SPS change scores (reflecting treatment outcomes) by the three family complexity components (predictors) are also demonstrated. The section ends with an illustration of the difference in SPS scores from baseline to short-term follow-up between low maternal RQI group and high maternal RQI group, a comparison selected because RQI emerged as the best predictor of outcome.

In total, 36 families participated in the behavioural intervention in the larger study were initially screened based on the selection criteria of the present study. Among these families, five dropped out early before the treatment phase. The data of one family (with two siblings) were incomplete, and therefore this family was not included in the final pool of data for the current analysis. Regarding the final 30

families who completed the treatment phase, two children were siblings in one family, and this resulted in a total of 31 children in the final data set. There were 11 single-parent families and 19 two-parent families. Among the 11 children from single-parent families, nine were living with the mother, one was with the grandmother, and one was living with the father. Therefore, only maternal data were available in the single-mother families, and only paternal data were collected from the single-father family. Furthermore, the RQI was administered only to the 19 two-parent families since it was not applicable to the solo-parent families. A family adversity composite score derived as discussed above in the method section, was calculated for each of the families, and a SPS composite score was computed for each of the children.

### **Quality of Data**

Among the 30 families, 29 mothers and 21 fathers completed the DASS-21 scale; 22 mothers and 16 fathers completed the PSQI scale; 17 mothers and 14 fathers completed the RQI scale at the pre-treatment stage. For post-treatment data, 23 mothers and 18 fathers completed the DASS-21 scale; 17 mothers and 11 fathers completed the PSQI scale; 10 mothers and 9 fathers completed the RQI scale. With regard to the SPS scores of the children, all 31 children provided pre-treatment and post-treatment SPS scores. There were 23 out of 31 children who completed up to the short-term follow-up phase and 21 children completed up to the long-term follow-up stage. Because of this, 23 SPS short-term follow-up scores and 21 SPS long-term follow-up scores were available.

To ensure the quality of the data, the data were screened and cleaned for missing data and outliers before further analysing the data. When missing data were a concern, if a small fraction of records were missing, mean scores were used to replace the missing scores. In cases with a larger fraction of the entry were missing, the data

were excluded from the analysis. For processing a statistical computation, the method of available data analysis (i.e., pairwise deletion) was used because, compared with using complete-case analysis, it allowed more cases to be retained in one analysis (Kwak & Kim, 2017). This method, however, resulted in the variation of sample sizes between variables used in the analysis. When outliers were a concern, two methods of computation, one with the outliers and one without the outliers, were used for processing the same analysis. Given that the outliers were also observed true values, the results of the computations with the outliers were retained in most of the cases unless the outliers distorted the relationship or effects to a great extent.

### **An Overview of Data Distributions**

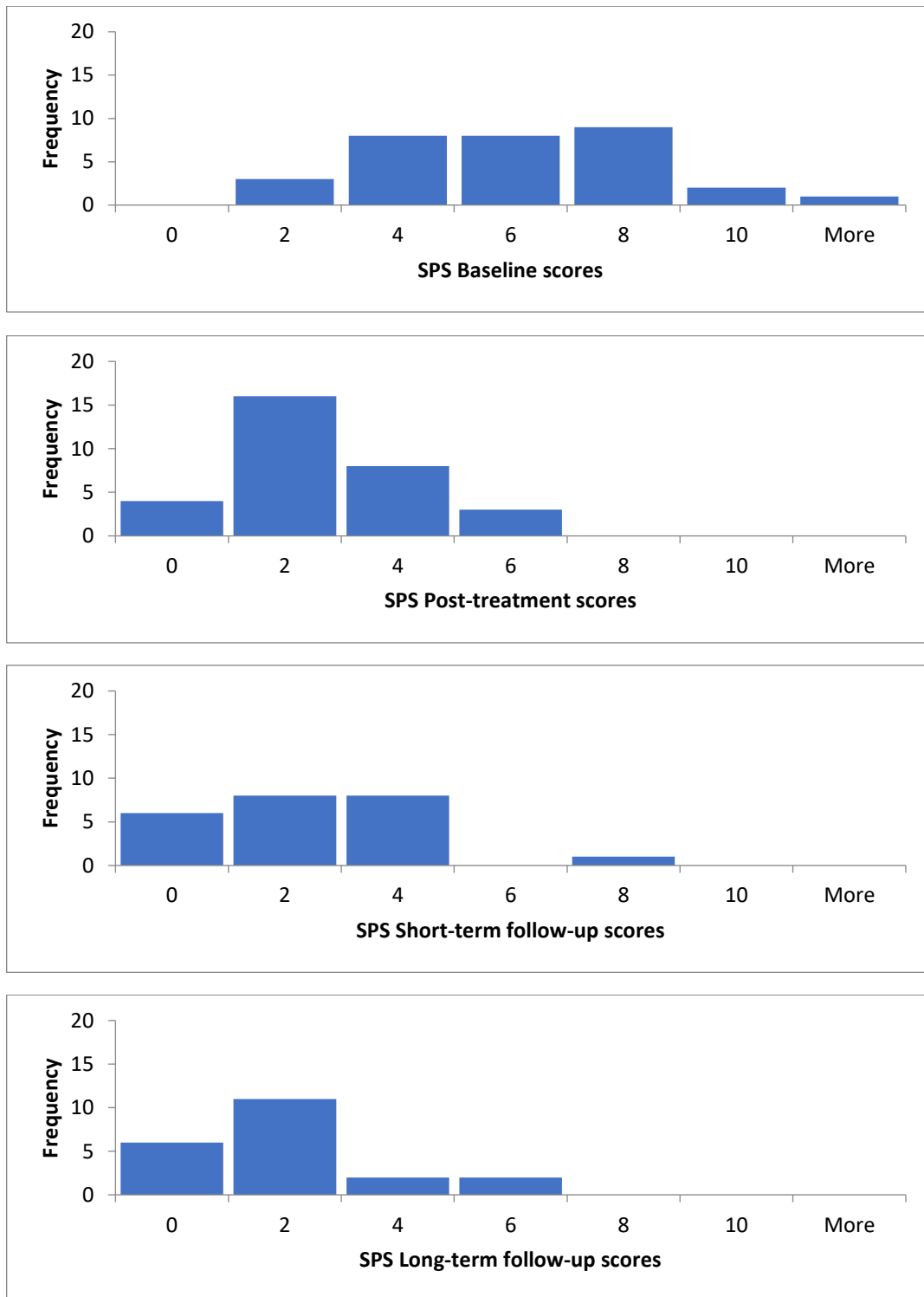
**Children sleep problem severity.** Figure 1 displays the frequency distributions of baseline, post-treatment, short-term follow-up and long-term follow-up SPS scores. The possible range of SPS score was from 0 to 23. The higher the score, the more serious the sleep problem severity. Frequency distributions enable the visual inspection of problems of non-normality in the data. If a skewness value is less than -1 or greater than 1, the distribution is highly skewed (Dancey & Reidy, 2004). If a kurtosis value is between -2 and +2, it is in the acceptable range of normal univariate distribution (George & Mallery, 2010).

***SPS baseline.*** The SPS scores at baseline phase showed a normal distribution with the majority of scores clustered between 4 and 8 (skewness = 0.59; kurtosis = 0.76).

***SPS post-treatment.*** The SPS scores at post-treatment phase also demonstrated a normal distribution with the majority of scores clustered between 2 and 4 (skewness = 0.79; kurtosis = 0.39).

***SPS short-term follow-up.*** The SPS scores at short-term follow-up phase shifted from a relatively normal distribution with the majority of scores being positively skewed, that is, scores distributed near the lower end of the scale (between 2 and 4), which suggested an overall reduction in sleep problem severity (skewness = 0.92; kurtosis = 0.73).

***SPS long-term follow-up.*** Long-term follow-up SPS displayed a positively skewed distribution with a majority of scores clustered between 0 and 2 denoting that most children had their sleep problem severity reduced (skewness = 1.38; kurtosis = 1.45).



*Figure 1.* Distribution of sleep problem severity (SPS) scores at baseline, post-treatment, short-term follow-up and long-term follow-up phases.

**Parental functioning measures.** The possible range of each DASS-21 subscores is from 0 to 42. The higher the score, the more the psychological distress. Figure 2 displays the frequency distributions of the paternal DASS-21 scores at pre-treatment and post-treatment phases. Figure 3 displays the maternal DASS-21 distributions at two time-points.

**DASS-21 fathers.** For DASS-depression subscale (DASS-D), a score of 10 to 13 denotes a mild level of depression. A score falls between 14 and 20 represents a moderate depression level. A score of 21 to 27 implies that the individual has a severe level of depression. A score of 28 or more indicates an extremely high level of depression. In the fathers, the depression scores demonstrated a relatively flat distribution at pre-treatment with scores quite evenly distributed across 0 to 16 that covered normal to moderate levels of depression, despite few high scores (skewness = 0.75; kurtosis = -0.16).

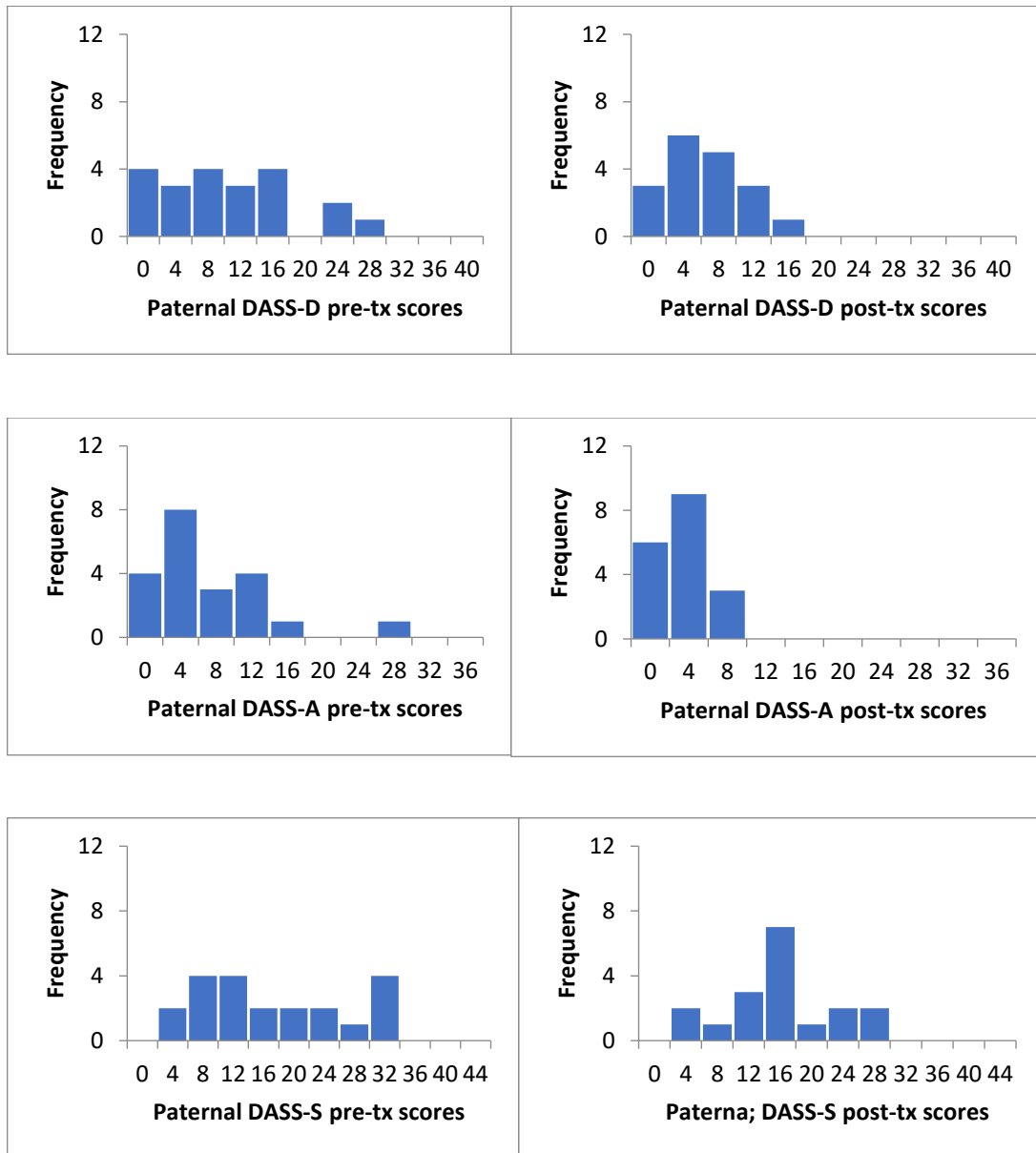
For DASS-anxiety subscale (DASS-A), a score of 8 to 9 denotes a mild level of anxiety. A score falls between 10 and 14 represents a moderate anxiety level. A score of 15 to 19 implies that the individual has a severe level of anxiety. A score of 20 or more indicates an extremely high level of anxiety. Regarding the anxiety profile at pre-treatment, the scores displayed a leptokurtic distribution with a high peak in score 4 denoting that most of the fathers had a normal anxiety level. The rest scores distributed across mild to moderate levels (skewness = 2.02; kurtosis = 5.56).

For DASS-stress subscale (DASS-S), a score of 15 to 18 denotes a mild level of stress. A score falls between 19 and 25 represents a moderate stress level. A score of 26 to 33 implies that the individual has a severe level of stress. A score of 34 or more indicates an extremely high level of stress. The stress distribution at pre-treatment was also in a flat fashion with scores quite evenly distributed across 4 and

32, covering normal to severe levels of stress (skewness = 0.35; kurtosis = -1.13).

Following treatment, in general, the frequency of high scores (e.g., score 32 or above) reduced or disappeared in the three DASS-21 subscales implying improvements in mood states in the fathers.





*Figure 2.* Distribution of paternal DASS-21 scores at pre-treatment and post-treatment phases.

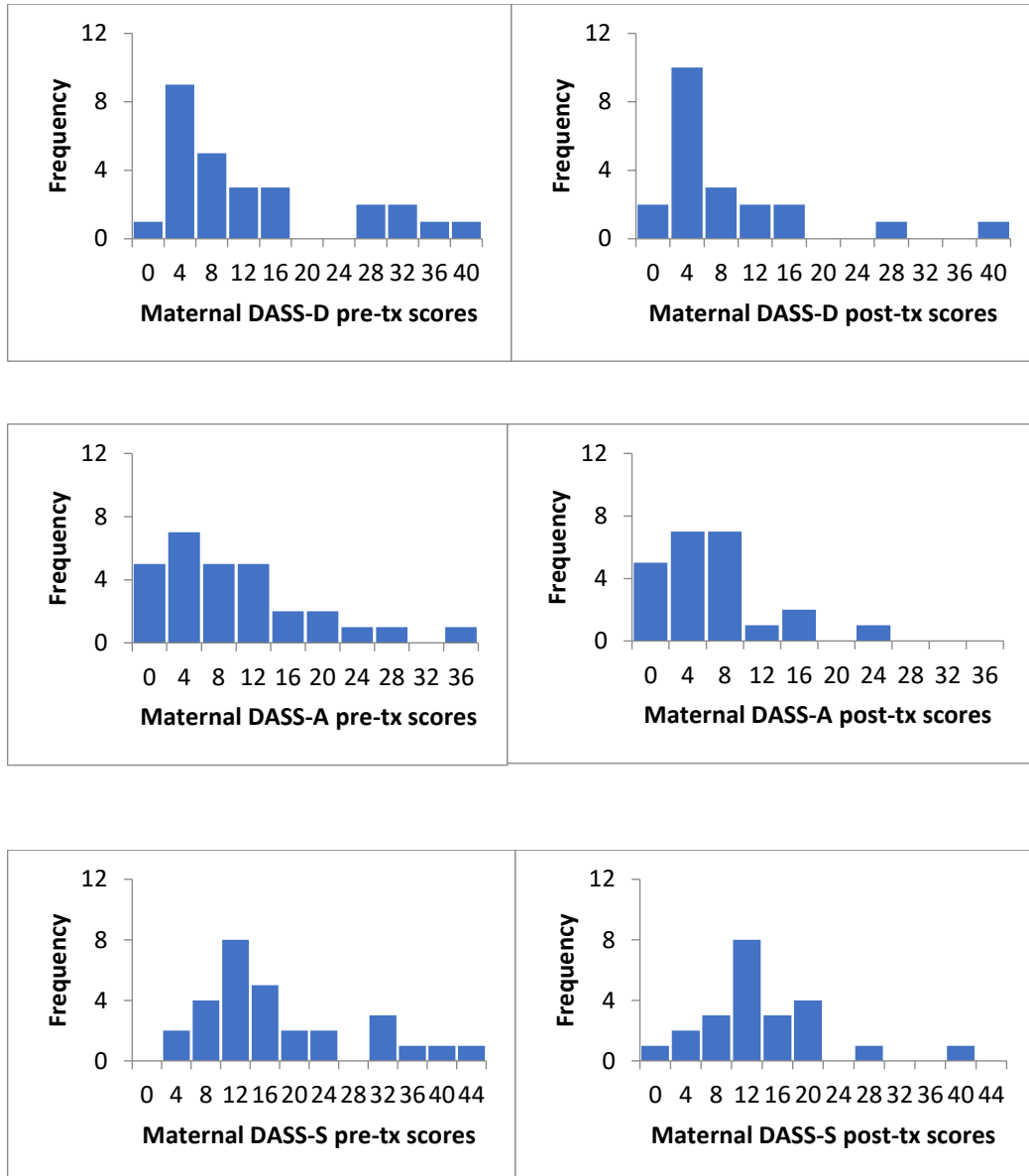
*Note.* DASS-D = DASS-depression; DASS-A = DASS-anxiety; DASS-S = DASS-stress; pre-tx = pre-treatment; post-tx = post-treatment.

**DASS-21 mothers.** In the mothers, the distribution of depression scores was positively skewed with the majority of scores clustered between 4 and 8, revealing that most of the mothers showed normal level of depressive symptoms despite the presence of several high scores (skewness = 1.25; kurtosis =0.33).

Similarly, the pre-treatment distribution of anxiety scores was also positively skewed with the majority of the scores clustered between 8 and 12 that covered normal to moderate ranges of anxiety level, in spite of several high scores (skewness=1.22; kurtosis=1.20).

For the stress subscale, the mothers demonstrated a relatively normal distribution profile with most scores spreading between 8 and 16 and covering normal to mild stress levels (skewness = 0.96; kurtosis= 0.13).

In general, following treatment, the frequency of high scores (e.g., score 32 or above) reduced in all three subscales of the DASS-21 as reflected in less extended tails of the histograms, representing an improvement in maternal mood states.



*Figure 3.* Distribution of maternal DASS-21 scores at pre-treatment and post-treatment phases.

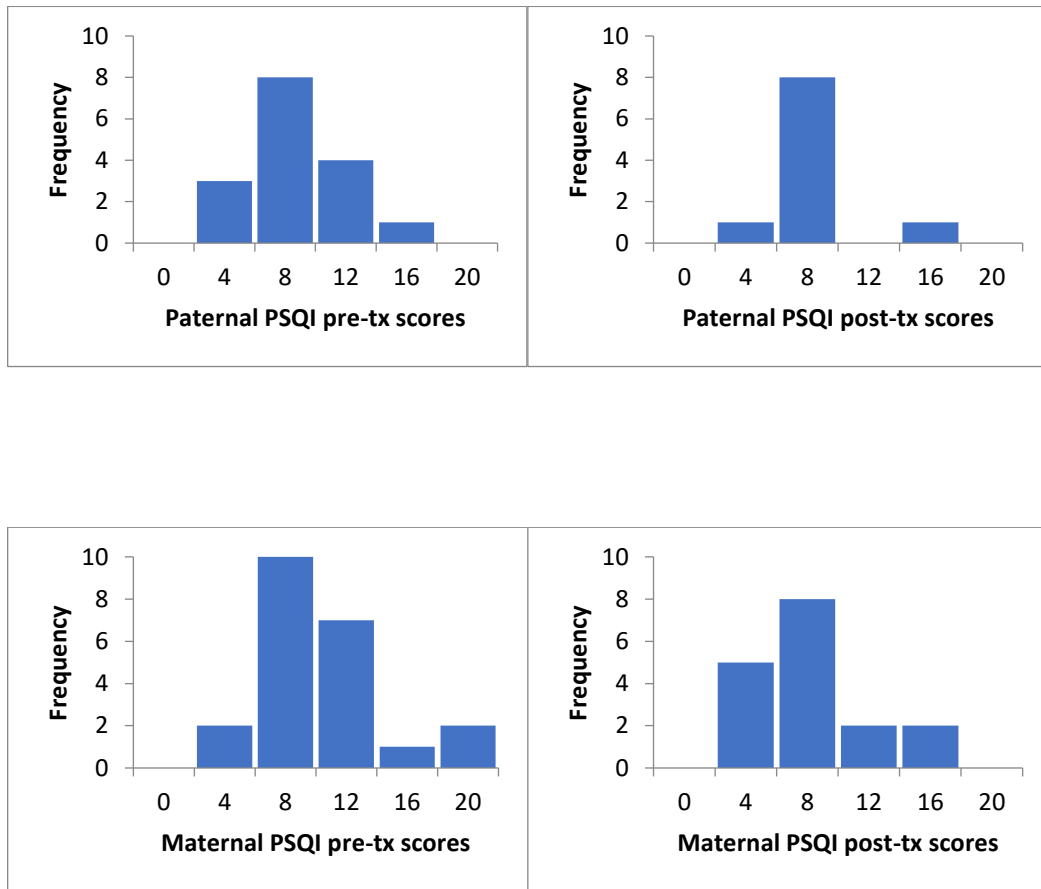
*Note.* DASS-D = DASS-depression; DASS-A = DASS-anxiety; DASS-S = DASS-stress; pre-tx = pre-treatment, post-tx = post-treatment.

**Parental PSQI.** The possible score range of the PSQI is between 0 and 21; the higher the score, the poorer the sleep quality. A total score of higher than 5 denotes poor sleep (Buysse et al., 1989). Figure 4 displays the distributions of paternal and maternal PSQI scores at pre-treatment and post-treatment phases.

The paternal PSQI scores showed a slightly positively skewed distribution at pre-treatment with the majority of scores clustered between 8 and 12 indicating an occurrence of poor sleep in the fathers (skewness = 1.38; kurtosis = 1.45).

The maternal PSQI scores at pre-treatment displayed a normal distribution with the majority of scores clustered between 8 and 12, which also indicated the poor sleep in the mothers (skewness = 0.66; kurtosis = 0.16).

Following treatment, the frequency of parents showed high scores (e.g., score 20 or more) were reduced denoting improvements in both the fathers' and the mothers' sleep quality.



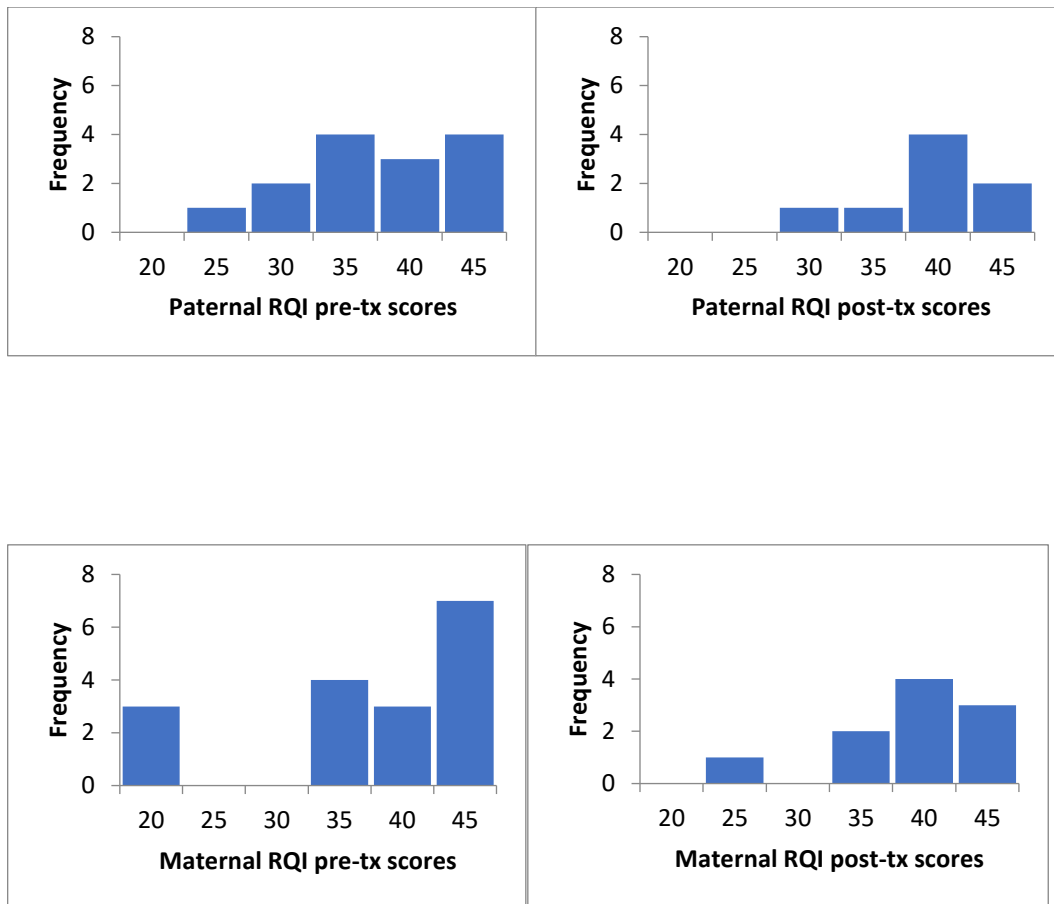
*Figure 4.* Distribution of paternal and maternal PSQI scores at pre-treatment and post-treatment phases.

*Note.* DASS-D = DASS-depression; DASS-A = DASS-anxiety; DASS-S = DASS-stress; pre-tx = pre-treatment; post-tx = post-treatment.

**Parental RQI.** The possible scores on the RQI range from 8 to 45; the higher the score, the better the marital satisfaction perceived by an individual. A score of less than 29 is indicative of relationship distress. Figure 5 displays the distributions of paternal and maternal RQI scores at pre-treatment and post-treatment phases.

The paternal RQI scores at pre-treatment phase were negatively skewed toward higher scores, with scores clustered between 35 and 45 (skewness = -1.02; kurtosis = -1.01). This indicated that fathers generally experienced a high level of overall relationship satisfaction. Following treatment, when compared to pre-treatment, fewer fathers had low scores that implied improvements in marital satisfaction in the fathers.

The maternal RQI scores were also negatively skewed to higher scores despite several low scores (skewness = -1.29; kurtosis = 0.90). Following treatment, when compared to pre-treatment, fewer mothers scored low in RQI; however, there were also fewer mothers that showed high scores. This suggested that some mothers felt less satisfied and some felt more satisfied in comparison to what they felt at the pre-treatment phase.



*Figure 5.* Distribution of paternal and maternal RQI scores at pre-treatment and post-treatment phases.

*Note.* DASS-D = DASS-depression; DASS-A = DASS-anxiety; DASS-S = DASS-stress; pre-tx = pre-treatment; post-tx = post-treatment.

## **Effect of Behavioural Sleep Treatment on Children's Sleep Problems**

**Descriptive statistics of SPS.** Table 4 provides the means, standard deviations, and medians of baseline, post-treatment, short-term follow-up, and long-term follow-up scores on the SPS of the 31 children.

Before treatment, the average sleep severity score was 5.80, and it dropped to 2.06 after treatment. The mean sleep severity score further reduced to 1.96 in the short-term follow-up phase, and eventually, it dropped to 1.57 in the long-term follow-up phase. This continuous reduction trend indicated that the sleep problems of the children were improving from pre-treatment to post-treatment and the gains lasted to the long-term follow-up phase.

To quantify change, mean change scores were computed for the differences in SPS scores between the baseline phase (i.e., pre-treatment) and each of the three phases after treatment. By comparing the baseline scores and post-treatment scores, the mean change score was 3.74. The mean change score was 3.65 for those 23 children who remained in the study in the short-term follow-up phase. Twenty-one children who reached the long-term follow-up phase produced a mean change score of 4.19. In general, the children showed improvement in the post-treatment phase, and the improvements continued in two follow-up phases. Comparatively, more gains were observed between baseline and long-term follow-up phases.



Table 4

*Means and Standard Deviations for Children's SPS Scores*

Child outcomes	N	Mean	SD	Median	Range	Min	Max
SPS BL	31	5.80	2.52	6.00	11	2	13
SPS Post-tx	31	2.06	1.53	2.00	6	0	6
SPS STFU	23	1.96	1.85	1.00	7	0	7
SPS LTFU	21	1.57	1.69	1.00	6	0	6
SPS change score : SPS BL vs SPS Post-tx	31	3.74	2.77	3.00	13	-1	12
SPS change score: SPS BL vs SPS STFU	23	3.65	2.87	4.00	10	-1	9
SPS change score: SPS BL vs SPS LTFU	21	4.19	2.94	4.00	12	-2	10

*Note.* N= Participants; SD =Standard Deviation; Min = minimum score, Max= Maximum score; SPS= sleep severity problem; BL=baseline, Post-tx = post-treatment; STFU = short-term follow-up; LTFU= long-term follow-up

**Primary treatment effects – child sleep.** Research Question 1 aimed at investigating whether the behavioural treatment would alter the sleep problem severity of the children to a clinically significant level. Modified Brinley's plots were used to illustrate the changes in SPS scores of each child over time in order to identify any systematic effects of the sleep intervention. The general interpretation of the modified Brinley's plot is presented in Figure 6. The changes in SPS scores are presented in three modified Brinley's plots to demonstrate changes of each child from (1) baseline to post-intervention (see Figure 7), (2) baseline to short-term follow-up (see Figure 8), and (3) baseline to long-term follow-up (see Figure 9).

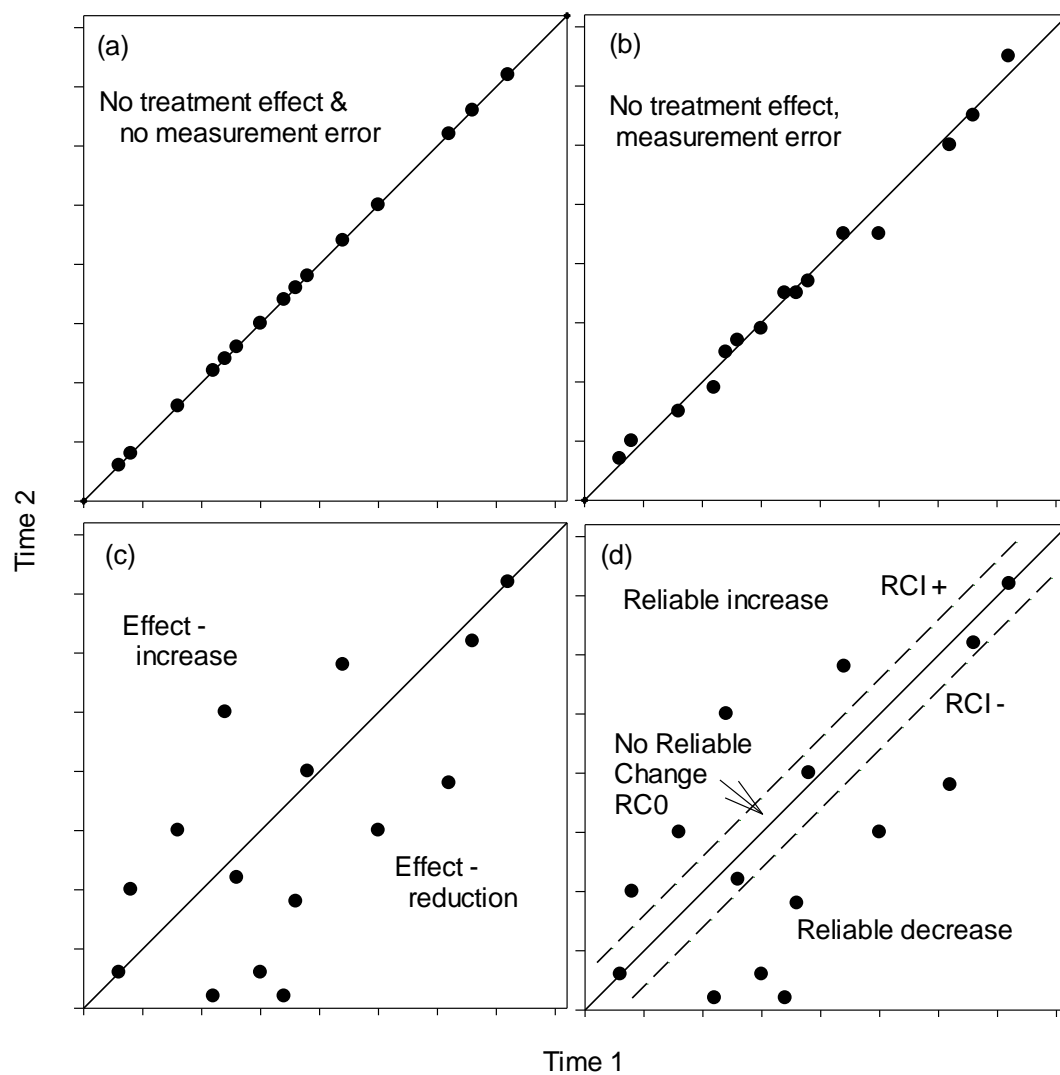
After the visual inspection of therapeutic changes between any two time points on the modified Brinley's plots, the Wilcoxon signed-rank test was used to judge the statistical significance of the change from one time point to the other.

Two Effect Sizes (ES) were computed in the present analysis to communicate the practical significance of the results. The first one is Cohen's  $d_{av}$  (Lakens, 2013). The second one is the Probability of Superiority (PS) which is also referred to as the Common Language Effect Size (McGraw & Wong, 1992). Cohen's  $d$  is a general term that includes different subtypes of effects sizes (Lakens, 2013). The present study was a within-subjects design, and so the effect size was calculated by using the average standard deviation of two time-points as the standardiser. Lakens (2013) labelled this type of effect size as Cohen's  $d_{av}$  but because only  $d_{av}$  is used in the present study, the subscription is omitted hereafter for simplicity in presentation.

In the following illustrations, a negative value of  $d$  indicates that the effect decreases the mean of that variable at post-treatment, and a positive value of  $d$  indicates the effect increases the mean of that variable at post-treatment. In the present analysis, the interpretation of the effect sizes followed the suggestion of Cohen (1988)

that  $d = 0.2$  was considered a small effect size,  $d = 0.5$  represented a medium effect size, and  $d = 0.8$  denoted a large effect size.

McGraw and Wong (1992) have proposed the Probability of Superiority that converts an effect size into a probability. The probability are reported in percentage terms, which indicates that, for any randomly selected participant, the score at Time 1 (e.g., baseline) is clinically different from the score in Time 2 (e.g., post-treatment) (Lakens, 2013; McGraw & Wong, 1992). The PS was calculated by using the software of Lakens (2013).



*Figure 6: Modified Brinley plots interpretation. Plot (a) to (c) display the general interpretations of a modified Brinley plot. Each data point denotes a score of time point 1 against time point 2 of a participant. The 45-degree diagonal line represents no difference/no treatment response. Plot (d) presents the upper and lower boundaries of Reliable Change. The portion beyond the upper dashed line above the 45-degree line belongs to the region of reliable increase (RC+). The portion underneath the dashed line below the 45-degree line belongs to the region of reliable decrease (RC-). The portion between two dashed lines is the region of no reliable change (RC0). (From Blampied, 2017, with permission).*

***Modified Brinley plots interpretation.*** As seen in Figure 6, each participant's data is displayed as a coordinate pair on a scatter plot with the first time-point score (e.g., SPS BL) plotted on the X-axis and the second time point score (e.g., SPS STFU) on the Y-axis. The 45-degree diagonal line represents a line of no change in scores between two time points (Figure 6a). Treatment response, if any, is denoted by data deviations from the 45-degree diagonal line. No systematic difference between two time points is denoted by those data points locating along or close to the 45-degree diagonal line (i.e.  $X \approx Y$ ; Figure 6b). Data points that situate away from the diagonal line (irrespective of whether above or below) show there are systematic differences due to time/treatment (Figure 6c; Blampied, 2017). When a lower score represents a clinical improvement, data points that fall below the diagonal line indicate improvement and those above the line indicate deterioration (Figure 6c). When a higher score represents a clinical improvement, the reverse explanation applies.

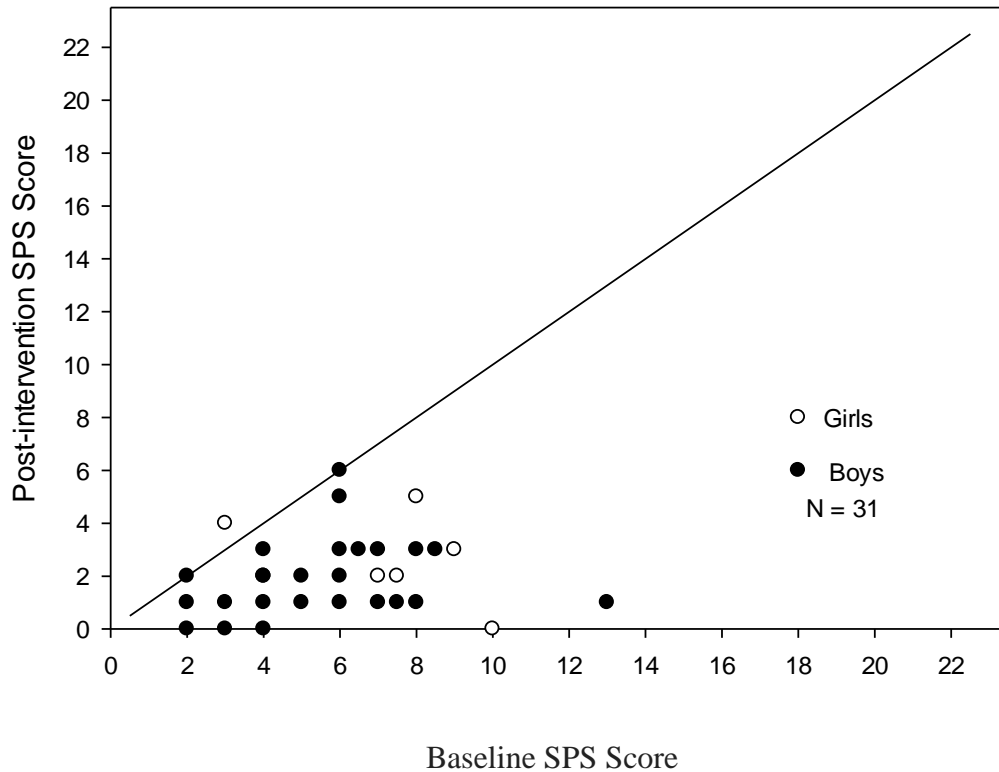


Figure 7. Changes in SPS scores at baseline phase against the post-intervention phase.

*Note.* The data points that have the same values would overlap. The values were jittered by adding 0.5, so they move sideways and up a little on the chart. This enabled overlapping points to be displayed.

*SPS baseline vs SPS post-treatment.* As seen in Figure 7, all 31 children had their total SPS scores scored above 0 at the baseline phase. Of these children, one experienced a small increase in sleep problem severity while 28 showed a reduction in such severity at the post-treatment phase. Two children demonstrated no change between two time points. The results of Wilcoxon signed-rank test showed that the sleep intervention produced a statistically significant change in sleep problem severity in the children ( $Z = -4.66, p < .001$ ). The SPS baseline score (median = 6.00) was significantly higher than the SPS post-treatment score (median = 2.00). Lower SPS

scores represented reduced sleep problem severity. The result of the Probability of Superiority indicated that the chance that a child experienced higher sleep problem severity at baseline than at post-treatment was 89.80%.

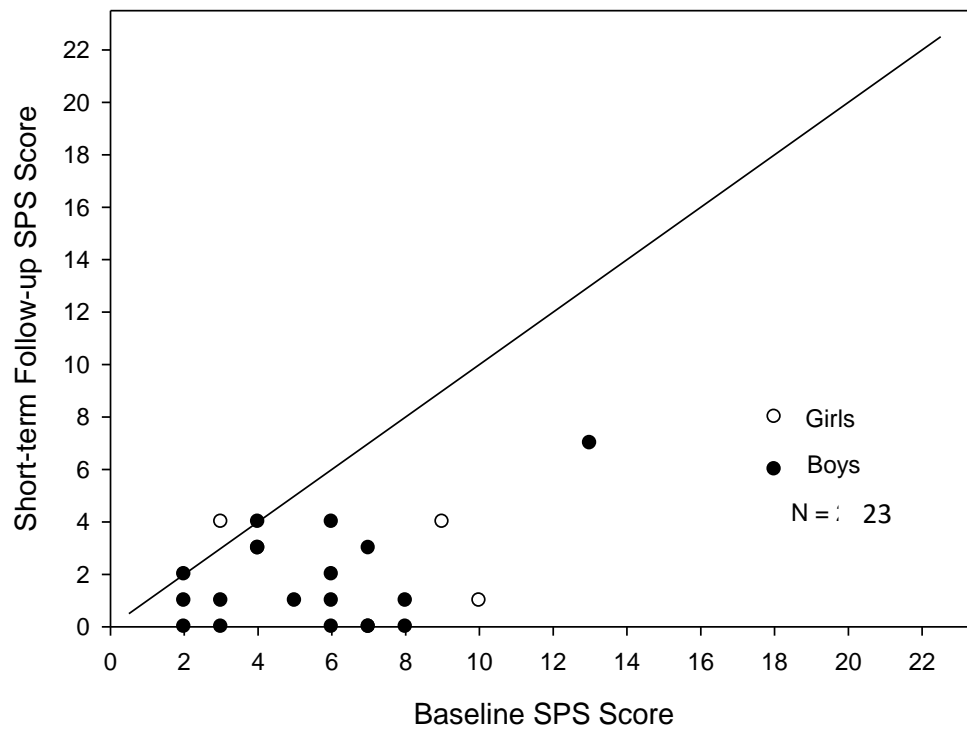


Figure 8. Changes of SPS scores at baseline phase against short-term follow-up phase.

Note The caption is otherwise the same as for Figure 7.

*SPS baseline vs SPS short-term follow-up.* Figure 8 shows the data of all 23 children who completed the short-term follow-up phase. Twenty of these children showed improvements in sleep problem severity while two children did not demonstrate any gains at short-term follow-up phase. One child had a small increase in total SPS score following short-term follow-up. The results of Wilcoxon signed-rank test also revealed a statistically significant improvement in the children's treatment outcomes ( $Z = -3.92, p < .001$ ). The SPS baseline score (median = 6.00) was significantly higher than the SPS short-term follow-up score (median = 1.00). The result of the Probability of Superiority indicated that the chance that a child



experienced higher sleep problem severity at baseline than at short-term follow-up was 89.07%.

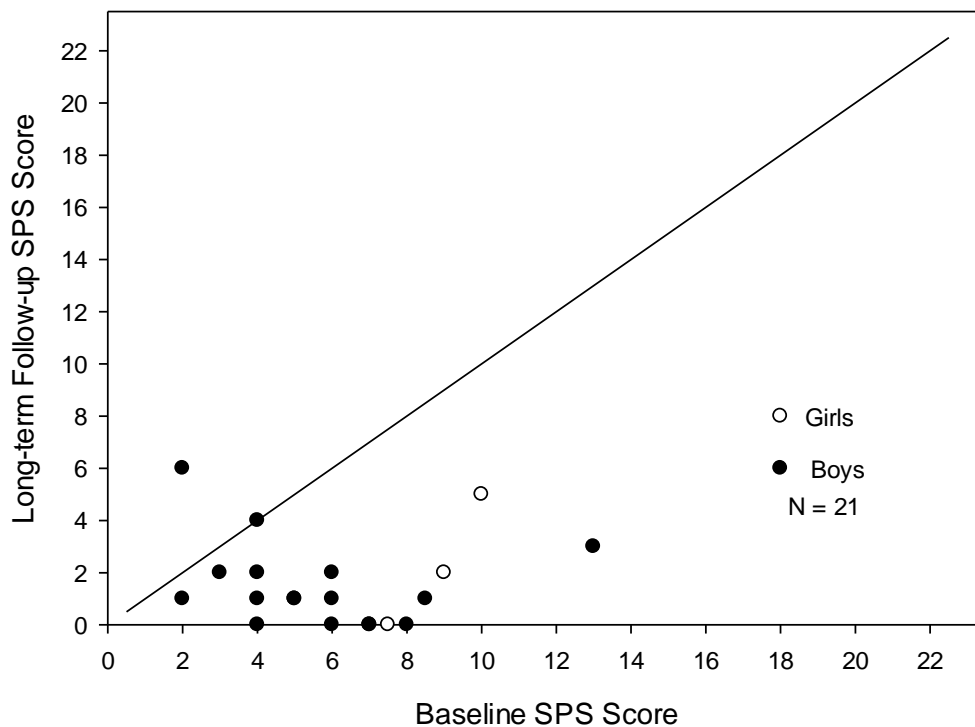


Figure 9. Changes of SPS scores at baseline phase against long-term follow-up phase.

Note The caption is otherwise the same as for Figure 7.

*SPS baseline vs SPS long-term follow-up.* Figure 9 illustrates the SPS scores of 21 children who finished the long-term follow-up phase. Among them, 19 children showed improvement in sleep problem severity while one child remained no change in comparison with the baseline phase. The severity of the sleep problem of the children demonstrated elevation following long-term follow-up. The results of Wilcoxon signed-rank test denoted a statistically significant improvement in the children's treatment outcomes ( $Z = -3.78, p < .001$ ). The SPS baseline score (median = 6.00) was significantly higher than the SPS long-term follow-up score (median = 1.00). As indicated by results of the Probability of Superiority, the chance that a child

experienced higher sleep problem severity at baseline than at short-term follow-up was 91.77%.

In summary, when compared to baseline, most children's sleep improved to a clinically significant degree following the intervention, and they displayed further improvement at short-term follow-up phase, with these gains sustained at the long-term follow-up phase. The behavioural intervention altered the child sleep to a clinically significant degree. This answered Research Question 1. A summary of Wilcoxon Signed-rank test results and Probability of Superiority of SPS scores across phases is presented in Table 5.

Table 5

*Summary of Wilcoxon Signed-rank Test Results and Probability of Superiority of SPS Scores Across Phases*

	BL		Post-tx		STFU		LTFU		Z	p	PS (%)
	N	Median	N	Median	N	Median	N	Median			
SPS scores	31	6.00	31	2.00					-4.66**	.001	89.80
	31	6.00			23	1.00			-3.92**	.001	89.07
	31	6.00					21	1.00	-3.78**	.001	91.77

*Note.* N = Participants; SD =Standard Deviation; Z = Wilcoxon z-value; PS = Probability of Superiority; BL=baseline;

Post-tx = post-treatment; STFU = short-term follow-up; LTFU = long-term follow-up; \*\* $p < .001$

**Secondary treatment outcomes – parental functioning.** Table 6 provides the means, standard deviations and *t*-test results for pre-and post-treatment scores on the DASS-21, PSQI, and RQI of the parents. The pre-treatment Family Adversity scores of the 30 families are also presented in Table 6. In the section below changes in group mean scores are presented.

**DASS-21 fathers.** For pre-treatment DASS-21 scores, in the fathers, the average stress subscore (mean =16.48) fell within the *mild* stress range (i.e., 15 to 18). The mean anxiety subscore (6.00) was close to the upper limit of the *normal* range (i.e., 0 to 7). The mean depression subscore (9.62) slightly exceeded the *normal* depression range (i.e., 0 to 9). Following treatment, paternal stress subscore slightly reduced to an average of 15.00; this remained within the *mild* range. The paternal anxiety subscore reduced to 2.67, which remained within the *normal* range. The paternal depression subscore dropped to 5.67 that fell within the *normal* range. For the comparisons of DASS-21 subscores between pre-treatment and post-treatment phases, the *t*-test results revealed that the paternal anxiety level showed a statistically significant reduction at post-treatment,  $t(17) = -5.72, p < .001, d = -0.67$ . The reduction of depression level at post-treatment also reached statistical significance,  $t(17) = -4.01, p < .001, d = -0.60$ . The two effect sizes are considered to be at a medium level (Cohen, 1988). The paternal stress level did not show a significant difference at the post-treatment time point when compared to pre-treatment.

**DASS-21 mothers.** For the mothers, the average subscores of stress (16.76), anxiety (8.90), and depression (11.59) fell within the *mild* ranges in the pre-treatment phase. After treatment, the maternal stress subscore dropped to an average of 13.21, which fell within the *normal* range. The maternal anxiety subscore decreased to an average of 5.65, and this belonged to the *normal* range. The maternal depression

subscore reduced to an average of 8.00, which was in the *normal* range. The *t*-test results indicated that the mothers experienced a significant decrease in anxiety level at the post-treatment phase,  $t(22) = -2.59, p = .017$ . The effect size for this analysis was small to moderate ( $d = -0.42$ ). The changes in stress level were found to be statistically non-significant. The difference in depression also did not reach statistical significance regardless of an improvement from mild to normal levels.

**PSQI.** For pre-treatment PSQI scores, both paternal (mean = 7.13) and average maternal scores (mean = 9.27) were within the clinical range (i.e., score  $\geq 5$ ). Following treatment, paternal score slightly increased to 7.50 while maternal score reduced to 7.12, although both scores remained within the clinical range. The deterioration in sleep quality of the fathers was very small, and did not reach statistical significance. The sleep improvement of the mothers was statistically significant, and yielded a medium effect size,  $t(16) = -2.17, p = .045, d = -0.54$ .

**RQI.** Before treatment, the average paternal RQI score was 35.43, and maternal RQI score was 35.12. Following treatment, the paternal score increased to 38.33, and maternal score increased to 36.80. All four scores did not fall within the *relationship distress* range (i.e., score  $\leq 29$ ). Although there were slight improvements in both paternal and maternal marital satisfaction ratings, the results of the *t*-test were found to be non-significant for both fathers' and mothers' scores.

In summary, following treatment, several secondary outcomes were evident in the parents, which included: (1) a reduction in paternal anxiety and depression levels, (2) a reduction in maternal anxiety level, and (3) an improvement in maternal sleep quality.

Table 6

*Summary of Means, Standard Deviations and t-test Results of 11 Parent/Family Variables Obtained at Pre-treatment*

Parent/Family variables	Pre			Post			<i>t</i>	<i>df</i>	<i>p</i>	95% CI	Cohen's <i>d</i>
	<i>N</i>	<i>Mean</i>	<i>SD</i>	<i>N</i>	<i>Mean</i>	<i>SD</i>					
Paternal DASS-D pre-tx	21	9.62	8.38	18	5.67	4.19	-4.01**	17	.001	[-6.03, -1.87]	-0.60
Paternal DASS-A pre-tx	21	6.00	6.54	18	2.67	2.47	-5.72**	17	.001	[-4.56, -2.10]	-0.67
Paternal DASS-S pre-tx	21	16.48	9.88	18	15.00	6.66	-0.94	17	.360	[-4.79, 1.84]	-0.21
Paternal DASS-Total pre-tx	21	31.90	22.16	18	23.33	10.91					
Maternal DASS-D pre-tx	29	11.59	11.37	23	8.00	9.40	-1.83	22	.081	[-7.65, 0.48]	-0.34
Maternal DASS-A pre-tx	29	8.90	8.83	23	5.65	6.02	-2.59*	22	.017	[-5.84, 0.64]	-0.42
Maternal DASS-S pre-tx	29	16.76	10.93	23	13.22	8.71	-1.95	22	.064	[-7.31, 0.22]	-0.36
Maternal DASS-Total pre-tx	29	18.62	13.97	23	26.87	21.49					
Paternal PSQI pre-tx	16	7.13	3.38	11	7.50	2.87	0.43	10	.674	[-1.55, 2.30]	0.12
Maternal PSQI pre-tx	22	9.27	3.82	17	7.12	4.09	-2.17*	16	.045	[-4.26, 0.05]	-0.54
Paternal RQI pre-tx	14	35.43	6.79	9	38.33	5.61	1.55	8	.159	[-1.41, 7.22]	0.47
Maternal RQI pre-tx	17	35.12	10.80	10	36.80	6.12	0.87	9	.408	[-2.70, 6.06]	0.19
Family adversity	31	2.71	1.40								

*Note.* N= Participants; SD =Standard Deviation; DASS-D = DASS-depression; DASS-A = DASS-anxiety; DASS-S = DASS-stress; DASS-Total = sum of DASS-depression ,DASS-anxiety, and DASS-stress subscores; pre-tx = pre-treatment; 95% CI = 95% confidence intervals; \* $p < .05$ , \*\* $p < .001$  (2-tailed).

## **Correlations between Pre-treatment Parent/Family Scores and Children's SPS Scores Across Phases**

In order to examine the relationships (1) among the 11 parent/family variables obtained at pre-treatment (i.e., DASS-21, PSQI, RQI and the family adversity scores), and (2) between these 11 parent/family variables and four child sleep problem variables (i.e., SPS at baseline, post-treatment, short-term and long-term follow-up), Spearman's rank order correlation coefficients were computed to determine the relationships between pre-treatment scores of 11 parent/family variables and the four SPS scores. These correlations are presented in Table 7.

For relationships among parent and family variables, results showed fifteen instances of statistically significant correlations (i.e., correlations  $> 0$  and  $p$ -value  $< .05$ ) among the fifteen variables. These fifteen cases of correlations were large enough to reject the null hypothesis that true  $r = 0$ . As expected, the three DASS-21 subscores of the fathers were moderately positively correlated among each other ( $r = .63$  to  $.76$ ). A similar pattern was observed among the three DASS-21 subscores of the mothers ( $r = .49$  to  $.67$ ). The DASS-depression and DASS-stress subscores of the fathers were also moderately positively correlated with their own PSQI score indicating that high ratings of paternal emotional disturbance were correlated with high ratings of paternal sleep difficulties ( $r = .55$  to  $.63$ ). The three DASS-21 subscores of the mothers were also positively correlated with the mothers' PSQI score ( $r = .63$  to  $.85$ ), with the DASS-anxiety subscore demonstrating a relatively stronger relationship that denoted the more the anxiety experienced by the mothers, the poorer the sleep quality they had. The RQI scores of two parents were highly positively correlated to each other indicating that high ratings of paternal relationship satisfaction were correlated with high ratings of maternal relationship satisfaction ( $r = .82$ ). Finally, the family

adversity score was mildly positively correlated with the maternal DASS-depression and DASS-stress subscores, representing that high scores of family adversity were correlated with high scores of maternal depression and stress ( $r = .37$  to  $.56$ ). Other correlations were small and not significant.

For the relationship between parent/family variables and child variables, results indicated two instances of statistically significant correlations. A significant negative correlation was found between the children's SPS long-term follow-up score and the paternal ROI score representing that reduced severity of children's sleep problems at long-term follow-up was correlated with increased marital satisfaction perceived by the fathers( $r = -.73$ ). A similar trend was found between maternal RQI score and SPS long-term follow-up score, although this correlation did not reach statistical significance ( $r = .56, p = .061$ ). The second significant correlation was found between SPS short-term follow-up score and SPS long-term follow-up score indicating that reduced severity of children's sleep problems in short-term follow-up phase was correlated with reduced severity of their problems in long-term follow-up phase ( $r = .55$ ). Other correlations were not significant.

In summary, at the baseline phase, the fathers felt more depressed and stressed when they did not have a good sleep. The maternal mood states worsened when the mothers' sleep quality decreased. The mothers became more depressed and stressed when their family experienced more adversity. When the fathers perceived the marital relationship was satisfactory, the mothers also had a similar perception. If the children's sleep problem severity at short-term follow-up was low, the severity at long-term follow-up was likely to be low.



Table 7

*Spearman's Rank Order Correlations of 11 Pre-treatment Parent/family variables and Four Child SPS scores*

Variables	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15
1 Paternal DASS-D pre-tx	1.00	<b>.67**</b>	<b>.76**</b>	.35	.40	<b>.48*</b>	<b>.63**</b>	.35	-.12	-.15	.05	.11	-.10	-.24	.05
2 Paternal DASS-A pre-tx		1.00	<b>.63**</b>	.33	.14	-.06	.50	.02	-.04	.16	-.08	.00	-.09	-.04	-.26
3 Paternal DASS-S pre-tx			1.00	.22	.33	.34	<b>.55*</b>	.24	.05	-.11	-.02	.28	-.29	-.28	-.27
4 Maternal DASS-D Mum pre-tx				1.00	<b>.66**</b>	<b>.67**</b>	.27	<b>.63**</b>	.11	-.11	<b>.37*</b>	-.11	.04	-.28	.09
5 Maternal DASS-A Mum pre-tx					1.00	<b>.49**</b>	.30	<b>.85**</b>	-.03	-.38	.19	.09	-.13	-.42	.01
6 Maternal DASS-S Mum pre-tx						1.00	.23	<b>.70**</b>	-.13	-.32	<b>.56**</b>	-.20	-.04	-.31	.20
7 Paternal PSQI pre-tx							1.00	.36	.13	.30	-.39	.30	-.02	-.09	-.06
8 Maternal PSQI pre-tx								1.00	-.35	-.42	.29	-.05	.04	-.44	.16
9 Paternal RQI pre-tx									1.00	<b>.82**</b>	-.45	.11	-.36	-.21	<b>-.73*</b>
10 Maternal RQI pre-tx										1.00	-.32	.05	.05	.11	-.56
11 Family adversity score											1.00	-.20	-.23	-.08	.03
12 SPS BL												1.00	.21	.07	.07
13 SPS Post-tx													1.00	.288	.33
14 SPS STFU														1.00	<b>.55*</b>
15 SPS LTFU															1.00

*Note.* DASS-D = DASS-depression; DASS-A = DASS-anxiety; DASS-S = DASS-stress; pre-tx = pre-treatment; BL = baseline; Post-tx = post-treatment; STFU = short-term follow-up; LTFU = long-term follow-up; correlations printed in bold are significant; \* $p < .05$ , \*\* $p < .001$  (2-tailed).

### **Post-treatment Parent/Family Scores and Children's SPS Changes**

Table 8 illustrates the relationships between post-treatment parent scores, treatment acceptability scores and three children SPS change scores, that is, the SPS changes between (1) baseline and post-treatment, (2) post-treatment and short-term follow-up, and (3) short-term follow-up and long-term follow-up. Spearman's rank order correlation coefficients were computed to determine the directions and strengths of correlations.

For relationships between parent and children variables, results showed six instances of statistically significant correlation. At post-treatment, not surprisingly, the maternal PSQI score was moderately negatively correlated with the change in SPS from baseline to post-treatment ( $r = -.59$ ). At long-term follow-up, a similar pattern was found; the PSQI score of the mothers was moderately negatively correlated with the change in SPS score from baseline to long-term follow-up ( $r = -.65$ ). These two significant correlations indicated that the bigger the improvement the child demonstrated at post-treatment and long-term follow-up phases, the better the sleep quality the mother had. At short-term follow-up, however, an interesting pattern was observed in the positive correlation between maternal anxiety level and the change in SPS scores representing that the more gains the child showed at short-term follow-up, the more anxiety the mother experienced ( $r = .59$ ). The three SPS change scores were positively correlated with each other strongly ( $r = .78$  to  $.89$ ). These three significant correlations were in the expected direction denoting that more gains at post-treatment was correlated with more gains in the two follow-up time points.

Research Question 4 aimed at examining if there was any relationship between treatment acceptability and treatment outcomes. The parental TARF-R ratings were found not correlated with the SPS change scores. Neither paternal nor maternal

TARF-R scores were correlated with the parent/family variables. However, unsurprisingly, the paternal TARF-R score was found moderately positively correlated with the maternal TARF-R score ( $r = .62$ ). Other comparisons were found not significant.

In summary, following treatment, when the children's sleep showed more improvement, maternal sleep quality also improved more. This phenomenon replicated in the long-term follow-up phase. Interestingly, in the short-term follow-up phase, the mothers manifested more anxiety whereas the children showed more improvement. When a child had more gains at post-treatment, he/she was more likely to have more gains at short-term and long-term follow-up phases. When the fathers' treatment acceptability ratings were high, the mothers also rated high in the TARF-R. Nevertheless, treatment acceptability was found not correlated with any of the parent/family variables or the child variables.

Table 8

*Spearman's Rank Order Correlations of Post-treatment Parent Scores, Three Child SPS change scores and Parental TARF-R scores*

Variables	SPS change scores			TARF-R Dad	TARF-R Mum
	SPS BL vs SPS Post-tx	SPS BL vs SPS STFU	SPS BL vs SPS LTFU		
Paternal DASS-D post-tx	-.35	-.51	-.31	.40	-.12
Paternal DASS-A post-tx	-.00	-.14	.16	-.37	-.33
Paternal DASS-S post-tx	.13	.16	.27	-.02	.09
Maternal DASS-D post-tx	-.05	.04	.02	.38	-.01
Maternal DASS-A post-tx	.41	<b>.59*</b>	.45	.18	.18
Maternal DASS-S post-tx	-.31	-.27	-.31	.39	-.32
Paternal PSQI post-tx	-.48	-.39	-.41	.49	.03
Maternal PSQI post-tx	<b>-.59*</b>	-.35	<b>-.65**</b>	.59	.00
Paternal RQI post-tx	-.02	.20	.41	-.16	.09
Maternal RQI post-tx	.42	.27	.57	.03	-.15
SPS change score : SPS BL vs SPS Post-tx	1.00	<b>.78**</b>	<b>.89**</b>	-.53	-.28
SPS change score: SPS BL vs SPS STFU		1.00	<b>.89**</b>	-.45	.11
SPS change score: SPS BL vs SPS LTFU			1.00	-.59	-.18
Paternal TARF-R				1.00	<b>.62*</b>
Maternal TARF-R					1.00

*Note.* DASS-D = DASS-depression; DASS-A = DASS-anxiety; DASS-S = DASS-stress; post-tx = post-treatment; BL = baseline; STFU = short-term follow-up; LTFU = long-term follow-up; correlations printed in bold are significant; \* $p < .05$ , \*\* $p < .001$  (2-tailed).

## Exploratory Factor Analysis

To answer Research Question 2, principal components analysis was used in the exploratory factor analysis to identify whether there were one or more components of family/parental complexity underlying the 11 parent/family variables obtained at pre-treatment. In total, four factor analytic models were examined (i.e., Models 1, 2, 3, & 4).

Because the pre-treatment depression, anxiety, and stress subscores of DASS-21 were highly correlated ( $r = .63$  to  $.85$ ) with each other, we summed the three DASS-21 subscores of the fathers into the paternal DASS-21-total pre-treatment score, producing a composite DASS score measuring general psychological distress. Similar computation was also done for the mothers. Because of this, instead of six DASS-21 variables, two variables, paternal DASS-total pre-treatment and maternal DASS-total pre-treatment were included in the third and fourth factor analyses. Therefore, before the last two attempts (i.e., Models 3 & 4) were executed, 11 pre-treatment parent/family variables had been reduced to seven variables, namely, (1) paternal DASS-total pre-treatment, (2) paternal PSQI pre-treatment, (3) paternal RQI pre-treatment for the fathers, and (4) maternal DASS-total pre-treatment, (5) maternal PSQI pre-treatment, and (6) maternal RQI pre-treatment for the mothers, and (7) family adversity variable.

The family adversity variable was found moderately loaded (.49) on Factor 1 of Model 1 and not much on the other two factors. Therefore, the family adversity variable was excluded from the analysis in Model 2 and Model 4 to investigate the difference in the outcome of extracted factors.

The Kaiser-Meyer Olkin (KMO) measure of sampling adequacy was used to

confirm the appropriateness of using factor analysis. The results indicated the KMO statistic was greater than 0.50 and because of this factor analysis was considered as an appropriate technique for further analysis (Howard, 2016). A factor loading .45 was treated as a good rule of thumb for the minimum loading of a variable (Tabachnick & Fidell, 2007). This meant that any variable that carried a factor loading lower than .45 would be removed from the cluster of variables within an extracted factor. For a cross-loading variable (i.e., a variable that loads on more than 1 factor), the variable would be dropped from the analysis if it were strongly loaded ( $r \geq .50$ ) to more than one extracted factors (Costello & Osborne, 2005). In this analysis, no variable loaded strongly ( $r \geq .05$ ) on more than one extracted factor in any of these four models and, therefore, all 11 variables were retained in the analysis.

For the cluster of variables that strongly loaded on an extracted factor to be labelled as a new composite factor, there should be at least three variables (Tabachnick & Fidell, 2007). A factor with two variables was only considered reliable when the variables were highly correlated with each another ( $r > .70$ ) but fairly uncorrelated with other variables (Yong & Pearce, 2013). Based on the Kaiser Criterion, an extracted factor was retained if its eigenvalue was larger than 1 (Costello & Osborne, 2005). The scree plot was also used to plot each eigenvalue on a graph and determine which factor has to be retained. A factor with an eigenvalue before the elbow of the scree plot curve would be retained (Howard, 2016). In the case of cross-loadings, the loading on the factor with factor loading  $< 0.5$  would be ignored. Following the ‘eigenvalues-greater-than-one’ rule (Howard, 2016), three factors were extracted and retained in each model.

**Model 1.** For Model 1, factor analysis of the 11 parent/family variables revealed that three factors were sufficient to explain the underlying structure of family

complexity. The first factor was labelled Maternal and Family Wellbeing which included three maternal DASS-21 subscale, maternal PSQI, and family adversity variables. The second factor was named the Paternal Wellbeing that was composed of three paternal DASS-21 subscale and paternal PSQI variables. The third factor, Parental Relationship Quality, consisted of paternal RQI and maternal RQI variables. Although the third factor contained only two variables, these two variables highly correlated with each other ( $r > .70$ ), and so, this factor was identified as Parental Relationship Quality. The Maternal and Family Wellbeing factor had an eigenvalue of 3.84, and it accounted for 34.87% of the variance in the data. The Paternal Wellbeing factor had an eigenvalue of 2.50 and accounted for a further 22.68% of the variance. The eigenvalue for Parental Relationship Quality factor was 1.53 accounting for a further 13.93 % of the total variance. The total variance explained by this model was 71.48%. The percentage of variance explained of each factor, and factor loadings of each variable are presented in Table 9.

**Model 2.** The second attempt at exploratory factor analysis included the 10 parent variables only (i.e., without family adversity). Similar to Model 1, the results of Model 2 produced three factors. The first factor was labelled the Maternal Wellbeing including maternal DASS-21 total and maternal PSQI variables. The second factor was named the Paternal Wellbeing, and it was comprised of paternal DASS-21 total and paternal PSQI variables. The third factor was the Parental Relationship Quality consisting of paternal RQI and maternal RQI variables. In Model 2, Factor 1 had an eigenvalue of 3.72, and it accounted for 37.21% of the variance in the data. Factor 2 had an eigenvalue of 2.35 and accounted for a further 23.52% of the variance. The eigenvalue for Factor 3 was 1.52 accounting for a further 15.22 % of the total variance. The total variance explained by Model 2 was 75.95%.

**Model 3 and 4.** Similar to the results of the first two models, Model 3 and Model 4 also produced three factors each. Model 3 and Model 4 accounted for 75.57%, and 82.56% of total variance explained respectively. In Model 3, the Maternal and Family Wellbeing included maternal DASS-total, maternal PSQI and family adversity variables. Compared to Model 1, the factor loading of family adversity increased ( $r = .57$ ). In Model 4, each factor included two variables only. The two variables within each extracted factor were highly correlated and with increased factor loadings ( $r = .82$  to  $.93$ ) when compared to those in Model 2 ( $r = .71$  to  $.89$ ). The percentage of variance explained of Model 3 and Model 4 and factor loadings of each variable are presented in Table 10.

In summary, in response to Research Question 2, three common factors were found in the four attempts of exploratory factor analysis. The first factor was the Maternal and Family Wellbeing or Maternal Wellbeing (without family adversity). The second factor was the Paternal Wellbeing. The third factor was the Parental Relationship Quality. These three factors were considered to be the three components of family complexity for further multiple regression analyses.



Table 9

*Models 1 and 2 of Exploratory Factor Analysis with Varimax Rotation of 11 Pre-treatment Parent/Family Variables*

	Model 1			Model 2		
	Factor analysis			Factor analysis		
	Factor loadings	Variance explained	Eigen-values	Factor loadings	Variance explained	Eigen-values
Factor 1 :		34.87%	3.84		37.21%	3.72
Maternal DASS-D pre-tx	.88			.89		
Maternal DASS-A pre-tx	.87			.89		
Maternal DASS-S pre-tx	.85			.84		
Maternal PSQI pre-tx	.79			.80		
Family Adversity	.49			N/A		
Factor 2 :		22.68%	2.50		23.52%	2.35
Paternal DASS-S pre-tx	.89			.90		
Paternal DASS-D pre-tx	.88			.89		
Paternal PSQI pre-tx	.73			.72		
Paternal DASS-A pre-tx	.71			.71		
Factor 3 :		13.93%	1.53		15.22%	1.52
Paternal RQI pre-tx	.90			.90		
Maternal RQI pre-tx	.80			.82		
Total variance explained		71.48%			75.95%	

*Note.* DASS-D = DASS-depression; DASS-A = DASS-anxiety; DASS-S = DASS-stress; pre-tx = pre-treatment.

Table 10

*Models 3 and 4 of Exploratory Factor Analysis with Varimax Rotation of Seven Pre-treatment Parent/Family Variables*

	Model 3			Model 4		
	Factor analysis			Factor analysis		
	Factor loadings	Variance explained	Eigen-values	Factor loadings	Variance explained	Eigen-values
Factor 1 :		35.85%	2.51		39.03%	2.342
Maternal DASS-total pre-tx	.91			.92		
Maternal PSQI pre-tx	.84			.88		
Family Adversity	.57			N/A		
Factor 2 :		22.77%	1.59		24.00%	1.440
Paternal PSQI pre-tx	.90			.90		
Paternal DASS-total pre-tx	.80			.84		
Factor 3 :		16.96%	1.19		19.53%	1.172
Paternal RQI pre-tx	.94			.93		
Maternal RQI pre-tx	.80			.82		
Total variance explained		75.57%			82.56%	

*Note.* pre-tx = pre-treatment.

## Multiple Regression

To investigate whether family complexity components could predict treatment outcomes ( i.e., Research Question 3), multiple regression was used following the factor analysis. Among the four models resulted from factor analysis, Factor Model 2 accounted for the second highest variance and with the most interpretable structure, therefore, for the purpose of further analysis, the factors of Model 2 were used. A factor score was computed for each child for each factor of Model 2 (i.e., Maternal Wellbeing, Paternal Wellbeing, and Parental Relationship Quality).

Simultaneous multiple regression was carried out to investigate whether these three components of family complexity could significantly predict the child's treatment outcomes, i.e., the three SPS change scores, namely, SPS baseline vs SPS post-treatment, SPS baseline vs SPS short-term follow-up, and SPS baseline vs SPS long-term follow-up. Sociodemographic factors of the children (e.g., age, gender) and the parents (e.g., marital status) were entered into the regression analysis to test for any significant contribution to the treatment outcomes of the children.

The results indicated that sociodemographic factors such as age and gender of the children, and marital status of parents did not show significant contribution in any of the four regression models examined. The findings also revealed that only the prediction of SPS change score from baseline to short-term follow-up was significant across four regression models. The predictions at post-treatment and at long-term follow-up were found not significant in any of the four regression models computed.

Table 11 shows the results of regression computation of using Factor Model 2. The regression using factors of Model 2 indicated that this regression computation explained 37.82% of the variance and could significantly predict the SPS change

score, SPS baseline vs SPS short-term follow-up,  $F(3,19) = 3.86, p = .026$ . The Parental Relationship Quality variable contributed significantly to this model ( $\beta = .57, p = .008$ ). The other two components, Maternal Wellbeing ( $\beta = .33, p = \text{n.s.}$ ) and Paternal Wellbeing ( $\beta = .13, p = \text{n.s.}$ ), did not make a significant contribution. The final predictive model was:

$$\text{Treatment outcome SPS baseline vs SPS short-term follow-up} = 2.958 + (.57 * \text{Parental Relationship Quality})$$

Table 11

*Summary of Multiple Regression Analyses for Domain Variables of Family*

*Complexity in Predicting SPS Change Scores between Baseline and Short-term*

*Follow-up (n = 23)*

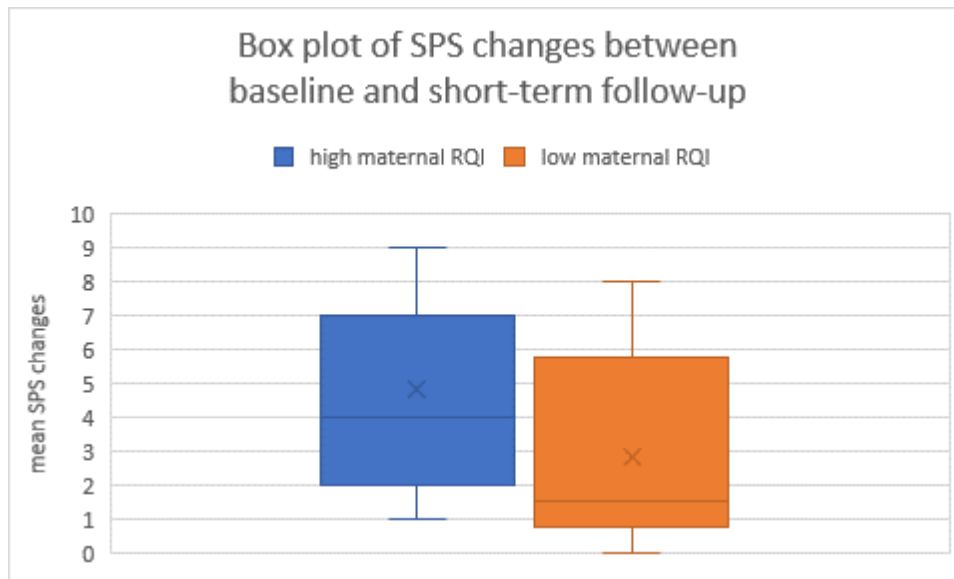
Factors in Model 2	Multiple Regression Analysis statistics				
	<i>B</i>	<i>SE B</i>	$\beta$	<i>t</i>	<i>p</i>
Maternal Wellbeing	0.93	.52	.33	1.78	.091
Paternal Wellbeing	0.36	.50	.13	0.72	.481
Parental Relationship Quality	2.26	.76	.57	2.97	.008
$R^2$	.38				
<i>F</i>	3.86				
<i>p</i>	.026				

### **The Effect of RQI on SPS: Comparison between Low RQI and High RQI Groups**

Thirteen children who had pre-treatment maternal RQI data available and completed the short-term follow-up phase were divided into two groups based on the median maternal RQI pre-treatment score (median RQI = 37). The box plots illustrate the differences in SPS change score (SPS baseline vs SPS short-term follow-up) between low maternal RQI group ( $n = 6$ ) and high maternal RQI group ( $n = 7$ ) (see Figure 5). The average SPS change score of the low maternal RQI group (mean SPS change score = 2.83) was lower than that of the high maternal RQI group (mean SPS change score = 4.86). The mean difference yielded a medium effect size ( $d = 0.64$ ) suggesting that two groups differed moderately in treatment outcome at short-term follow-up. The high maternal RQI group showed more treatment gains than the lower maternal RQI group at short-term follow-up.

Due to missing data of paternal RQI, the sample size was further reduced if we included the paternal RQI and divided the subsample into halves. Therefore, the analysis that divided the group based on paternal RQI or parental RQI is not presented here.

Among the three domain variables of family complexity, Parental Relationship was the only component that appeared to be a significant predictor of the treatment outcomes between baseline and short-term follow-up phases in the children. Maternal Wellbeing or Maternal and Family Wellbeing, and Paternal Wellbeing measured at pre-treatment did not predict the treatment outcomes substantially. These findings answered Research Question 3.



*Figure 10.* Box plot of high vs low maternal marital satisfaction subgroup comparison. Box plot displays the SPS changes between baseline and short-term follow-up of high maternal RQI (n = 7) and low maternal RQI (n = 6) groups

In summary, the intervention was found effective in reducing the sleep problem severity of the children, particularly at short-term follow-up phase (i.e., Research Question 1). Secondary outcomes included the reduction in paternal depression, paternal anxiety, and maternal anxiety levels, and improvement in maternal sleep quality. Based on the results of Model 2 in factor analysis, three components of family complexity, namely, Maternal Wellbeing, Paternal Wellbeing and Parental Relationship Quality, were extracted from the 11 pre-treatment parent/family variables (i.e., Research Question 2). Nonetheless, only the Parental Relationship Quality at pre-treatment could predict the treatment outcomes at short-term follow-up (i.e., Research Question 3). Treatment acceptability was found not correlated with the treatment response of the children (i.e., Research Question 4).

## **Chapter 5**

### **Discussion**

This study revealed that a parent-implemented, FBA informed, multi-component intervention was effective in ameliorating the sleep problems of the children with ASD. The reduction in sleep problem severity between baseline, intervention and follow-ups was found statistically significant. The findings also indicated that there were improvements in secondary outcomes such as paternal anxiety, paternal depression, maternal anxiety, and maternal sleep quality. According to the findings of the exploratory factor analysis, maternal wellbeing, paternal wellbeing and parental relationship quality appeared to be the components that resided underneath the 11 parent/family variables of family complexity, although parental relationship quality before treatment was found to be the only significant predictor of treatment outcomes. Interestingly, parental ratings of treatment acceptability were found to be unrelated to the treatment outcomes.

#### **Treatment Effectiveness on Child Sleep (Research Question 1)**

**Amelioration in child sleep problem severity.** The first research question investigated whether a FBA informed, home-based behavioural sleep treatment would result in a significant reduction in sleep problems for children with ASD. Among the 31 children included in the present study, 28 of them had reduced SPS scores between baseline and post-treatment with the group SPS score at baseline significantly higher than that at post-treatment indicating the amelioration of sleep problem severity following the 4- to 8-week sleep intervention. This result suggested an improvement in child sleep following treatment. The chance that a child had higher sleep problem severity at baseline than at post-treatment was 89.80%. Similarly, compared to baseline, reduction in sleep problem severity was also found at two follow-up phases. Twenty out of the 23 children who completed short-term follow-up



phase displayed a reduction in sleep problem severity with the group SPS score at baseline significantly higher than that at short-term follow-up (i.e., 4 to 6 weeks post-treatment). This result indicated a further attenuation of sleep problem severity at short-term follow-up. The chance that a child had higher sleep problem severity at baseline when compared to short-term follow-up was 89.07%. Nineteen of the 21 children who completed long-term follow-up phase reported a decrease in sleep problem severity with the group SPS score at baseline significantly higher than that at long-term follow-up (i.e., 10 to 12 weeks post-treatment). This result denoted a long-term improvement was evident in the children's sleep. The probability that a child showed higher sleep problem severity at pre-treatment than at long-term follow-up was 91.77%.

These findings are largely consistent with previous research which indicates that behavioural interventions are effective in reducing sleep problems in children with ASD (Christodulu & Durand, 2004; Durand, Gernert-Dott & Mapstone, 1996; Moore, 2004; Montgomery et al., 2004; Reed et al., 2009; Weiskop et al., 2005). The present results obtained at two follow-ups together with previous research that had also collected follow-up data suggest that treatment gains are likely to be maintained following the completion of treatment (Christodulu & Durand, 2004; Knight & Johnson, 2014; Maslow et al., 2016; Moon et al., 2011; Weiskop et al., 2005; Yu et al., 2015).

The present finding that FBA informed behavioural intervention was effective in treating sleep problems in children with ASD is consistent with previous research which has found that behavioural sleep treatment can reduce the duration and frequency of night wakings, sleep onset latency, increase total sleep time, and can result in the elimination of co-sleeping (Adkins, Mollow & Weiss, 2012; Austin, Gordon & O'Connell, 2013; Christodulu & Durand, 2004; DeLeon, Fisher & Marhefka, 2004.; Durand & Christodulu, 2004; Durand, Gernert-Dott & Mapstone, 1996; Johnson & Turner, 2013; Knight & Johnson, 2014; Loring,

Johnston, Gray, Goldman & Malow, 2016; Malow et al., 2014; Montgomery, Stores & Wiggs, 2004; Moon, Corkum & Smith, 2011; Moore, 2004; Piazza et al., 1998; Reed et al., 2009; Weiskop, Richdale, & Matthews, 2005; Yu et al., 2015). These findings are also supported by a number of systematic literature reviews which have concluded that a variety of antecedent- and consequence-based behavioural strategies are effective in addressing the sleep problems of children with ASD (Cuomo et al., 2017; Rigney et al., 2018; Turner & Johnson, 2012; Vriend et al., 2011). The behavioural strategies include antecedent-based strategies such as the use of sleep hygiene, faded bedtime and visual supports (Moon et al., 2010; Reed et al., 2009; Weiskop et al., 2001; 2005) and consequence-based strategies, such as standard or graduated extinction.

For the 31 children included in the present analysis, FBA outcomes were used to inform the selection of a mixture of antecedent-based and consequence-focused strategies in their treatment plans (McLay, France, Blampied, Danna, et al., 2017; McLay, France, Blampied, et al., 2017). These strategies included sleep hygiene modifications, faded bedtime, social stories and extinction-based procedures (McLay, France, Blampied, Danna, et al., 2017; McLay, France, Blampied, et al., 2017). It is likely that these procedures were effective in reducing the sleep onset latency, frequency of night waking, percentage of co-sleeping and intensity of bedtime resistance behaviours, and in increasing the total sleep time of the children in the present study; thus eventually, their sleep problem severity reduced (Durand et al., 1996; Montgomery et al., 2004; Moore, 2004; Reed et al., 2009; Weiskop et al., 2001; 2005). While it is difficult to determine the specific strategies that resulted in a reduction in sleep problem severity, the findings of this research provide evidence to support the use of FBA informed, parent-implemented interventions for sleep problems in children with ASD.

**Collateral benefits of improvement in child sleep.** Although investigating the secondary benefits of child sleep treatment was just an auxiliary task in the present study, the data collected allowed for an analysis of the collateral effect of improvement in child sleep on parental mood states (i.e., DASS-depression, DASS-anxiety and DASS-stress scores of the mothers and fathers), parental sleep (i.e., paternal and maternal PSQI scores), and parental relationship (i.e., paternal and maternal RQI scores). Significant improvements in paternal anxiety, maternal anxiety, paternal depression and maternal sleep quality were found when their children displayed sleep improvement at post-treatment. No apparent change was found in paternal sleep quality, parental stress and parental relationship. The maternal depressive symptoms displayed a decreased trend but not reach a statistically significant level.

***Parental mood states.*** Pre- and post- comparison of the DASS-anxiety scores revealed that, for the mothers, the change in maternal anxiety moved the average DASS-anxiety score from being in the *mild* range to the *normal* range. In the case of the fathers, the post-treatment average DASS-anxiety score remained in the *normal* range despite a significant reduction. Pre- and post- comparison of the DASS-depression scores indicated that in the fathers, the average paternal ratings of depression showed a change from the *mild* range to the *normal* range.

So far, no published ASD study, except one master thesis (Vivian, 2018), demonstrated the changes in parental functioning following a behavioural sleep intervention for children with ASD. Previous studies examining the relationship between child sleep and parental functioning have been limited to cross-sectional investigation (Chu & Richdale, 2009; Doo & Wing, 2006; Hoffman et al., 2006; Lopez-Wagner et al., 2008; Meltzer & Mindell, 2007). The present findings agreed with those found in two outcome studies that parental sleep and mood states improved following behavioural sleep treatments in TD children (Hall, Clauson & Carty, 2006; Hall et al., 2017). In general, the promising secondary

effects found in the present study added evidence to support the existing research findings that parents could also benefit from the child sleep intervention in children with ASD.

In general, the parents displayed improvements in mood states following treatment, particularly in anxiety and depression dimensions. There are a couple of plausible explanations for why improvement in child sleep may result in the uplifting of parental moods. It is likely that the experience of successfully implementing an in-home behavioural sleep programme may increase the sense of competence in the parents. Positive self-efficacy reinforced by the experience of success might enhance the mood states of the parents (Kuhn & Carter, 2006; Wiggs & Stores, 2001). It is also possible that improved child sleep enables the parents to have better and longer sleep. By that, improved parental sleep might contribute to elevated parental mood states (Meltzer & Mindell, 2007). Coincidentally, the mothers in the present study displayed improved sleep following treatment.

Although past studies which included TD children with sleep problems have demonstrated that improvement in child sleep may also reduce parental stress (Eckerberg, 2004; Lam et al., 2003; Reid et al., 1999; Scott & Richards, 1990), the present study did not replicate such findings. Interestingly, the findings of the present study aligned with previous research which has investigated parental stress in parents of children with developmental disabilities, and reported no apparent reduction in parental stress following intervention (Wiggs & Stores, 2001). It is possible that, although the parental stress levels did not show an apparent reduction at the 12-week follow-up in the present study, the parents may demonstrate decreased stress in a longer follow-up length (Eikeseth et al., 2015). Maternal stress was found reduced in 12 months following treatment (Elikeseth et al., 2015). Further investigation into any long-term change in parental mood post-treatment may shed some light on this interesting question.

***Parental sleep.*** Following treatment, there was a significant improvement in maternal sleep quality. This finding is consistent with previous research that has examined the relationship between parental sleep and child sleep in children with developmental disabilities, including children with ASD (Chu & Richdale, 2009; Meltzer, 2008; Polimeni et al., 2005), TD children, and children with intellectual deficits (Boergers et al., 2007; Robinson & Richdale, 2004; Wiggs & Stores, 2001). The parents in these studies who had disrupted sleep due to their child's sleep difficulties also reported improved sleep following treatment (Boergers et al., 2007; Chu & Richdale, 2009; Meltzer, 2008; Polimeni et al., 2005; Robinson & Richdale, 2004; Wiggs & Stores, 2001).

For the fathers, the pre- and post- comparison revealed there was no significant difference in paternal sleep quality following treatment. Few studies have examined the relationship between children's sleep problems and paternal sleep. Research that does exist suggested that the sleep of fathers of children with intellectual disabilities was negatively affected by their child's sleep disturbance; however such paternal sleep quality was found improved following treatment (Meltzer, 2008; Wiggs & Stores, 2001). In the present study, one possible explanation for the unaltered paternal sleep after treatment could be that there was a limited amount of data collected from fathers. Small number of available paternal cases ( $n = 11$ ) at post-treatment may be insufficient to yield a significant difference. Furthermore, about one-third of the families were solo parenting families in which the mother was the primary parent. It is likely that, as previously mentioned, usually, mothers are the primary caregivers of children with ASD. It is reasonable to infer that the maternal sleep quality is most affected by the child's sleep problems (Meltzer, 2008; Polimeni et al., 2005). Meltzer (2008) reported that maternal sleep quality was compromised due to the longer child sleep latency and frequent night waking in children with ASD. The findings of the present study

have suggested that improved sleep quality and the longer sleep duration of children with ASD might enable the mothers to sleep better and longer.

The results of the present study provide evidence of the positive effects of FBA informed behavioural treatment in reducing the sleep problem severity of the children with ASD. Furthermore, a reduction in sleep problems may have the secondary benefit of ameliorating the parental sleep quality and mood states, in particular, depression and anxiety symptoms. The findings that family members also benefited from improved child sleep have added some evidence to the ecological concept that members within a family system are interconnected and a change in one member can impact on the other members (Bronfenbrenner, 1985; Bowen, 1985).

### **Exploration of Family Complexity Components (Research Question 2)**

The second research question aimed to explore the underlying components of family complexity residing underneath the multiple pre-treatment parent/family that were used to define family complexity in the present study. The results of the present exploratory analysis suggested that there were three components or domains in family complexity, namely, maternal wellbeing, paternal wellbeing and parental relationship quality that resided underneath the 11 parent/family variables obtained at pre-treatment. The maternal wellbeing component included maternal mood states (i.e., depression, anxiety and stress) and maternal sleep variables. The paternal wellbeing component consisted of paternal mood states and paternal sleep variables. The parental relationship quality domain contained paternal marital satisfaction and maternal marital satisfaction. Kazdin and Whitley (2006) is the only study that has addressed case complexity in terms of parent/family characteristics, child dysfunction and barriers to participation. The findings of the present analysis shared the parental functioning and family relationship domains of family complexity used in Kazdin and Whitley (2006). However, the components found in the present study further

differentiated parental wellbeing into maternal and paternal wellbeing.

**Maternal wellbeing.** The maternal wellbeing component found in the present study included maternal mood states (i.e., stress, anxiety and depression) and maternal sleep measured prior to treatment. Kazdin and Whitley (2006) encompassed the measures of parental dysfunction and family relationships in the family complexity. The findings of the present analysis added to the understanding of family complexity that maternal sleep and maternal mood states were tied together to form one component. The present findings are in line with the existing literature that maternal sleep quality was correlated with maternal distress and stress in the mothers of TD children (Meltzer & Mindell, 2007) and of children with ASD (Chu & Richdale, 2009). One possible explanation is that disrupted sleep in the mothers may impair their ability to cope with the stress involved in parenting their child with ASD, which might eventually have negative impact (e.g., feeling of incompetence) on their mood states (Chu & Richdale, 2009). In mothers of children with ASD, fatigue was found positively correlated with maternal stress, anxiety and depression, and negatively correlated with parenting efficacy and satisfaction (Giallo, Wood, Jellett & Porter, 2013).

**Paternal wellbeing.** Inclusion of paternal data has been lagged in child sleep studies. Existing studies have not linked the paternal factor to child sleep problems (Counterline & Teti, 2010; Sadeh, Flint-Ofir, Tirosh, & Tikotzky, 2007). The paternal wellbeing component found in the present study consisted of paternal mood states and paternal sleep assessed at pre-treatment. This has been rarely mentioned in previous sleep studies in TD children (Bernier et al., 2013) and children with ASD (Meltzer, 2008). One possible explanation for paternal mood states with paternal sleep combined to form one component is that poor sleep may impair the day time functioning of the fathers, and eventually negatively affects their mood states (Meltzer, 2008). In a study focusing on the sleep disorders of adults, daytime sleepiness and sleep fragmentation were found related to depressive symptoms in males

(Basta et al., 2008).

**Parental relationship quality.** In the present study, the third component extracted from factor analysis was parental relationship quality, which has not been reported in sleep studies. While the family relationship in Kazdin and Whitley (2006) addressed the communication among family members, the parental relationship component found in the present study was more specifically focused on the relationship satisfaction between two parents. This expands the family relationship domain in the model of Kazdin and Whitley (2006). The parental relationship quality component found in the present study has suggested that the marital satisfaction of the mothers and the fathers are linked together. This relationship is aligned with the previous studies that the perceived support from one's spouse was linked to relationship satisfaction (Brobst, Clopton, & Hendrick, 2009).

### **Family Complexity Components Predicting Treatment Outcomes (Research Question 3)**

The third research question addressed whether the above-mentioned three components of family complexity (i.e., predictor variables) could predict sleep treatment outcomes in children with ASD. Parental relationship quality appeared to be the only predictor of treatment outcomes in the participants.

**Parental relationship quality.** When the data of all three components were simultaneously taken into account in the multiple regression analysis, parental relationship emerged as the only predictor of treatment responses in the children ( $n = 23$ ) and only at the short-term follow-up phase (i.e., 4 to 6 weeks post-treatment). This association was still evident after taking into account any possible confounders (e.g., marital status, SES, gender and age of the child) in the regression analysis. There were about 8 to 14 weeks in between pre-treatment parental data collection and short-term follow-up SPS scores consolidation. The present study is the first study which revealed that treatment outcomes can be predicted



by parental relationship quality in a short period of time following treatment conclusion. The present analysis also found that children from families with high maternal ratings of marital satisfaction demonstrated greater improvement in sleep problems than those from families of low maternal ratings of marital satisfaction. These findings are consistent with previous research which has mentioned that sleep problems are less likely to be resolved when marital problems occurred (Mindell et al., 2006; Narzisi, Muratori, Buscema, Calderoni & Grossi, 2015).

As mentioned above, the findings of the data within the duration in between pre-treatment parental data collection and short-term follow-up SPS scores consolidation disclosed that an about 8-to-14 week period of time might suffice to enable the parental relationship quality to produce an effect on treatment outcomes. Nevertheless, it seems that a shorter duration maybe not long enough to produce a similar positive effect because the prediction at post-treatment (i.e., 4 to 8 weeks) was not significant. It is difficult to determine how long the effect would become apparent because of the limited number of intervention studies that investigated the impact of marital relationship on child sleep. Some previous studies have indicated that behavioural strategies such as graduated extinction might take months to allow any remarkable change to be clearly demonstrated (Howlin 1984; Rodlier & Houton, 1984). The treatment outcomes obtained in the present study at the stage of post-treatment might be a bit too soon for the parental effect to be noticed. Future studies examining whether there exists any interaction effect between parental relationship quality and treatment duration on treatment outcomes might increase the understanding of the underlying mechanism.

The prediction at long-term follow-up (i.e., 10 to 12 weeks post-treatment) was also not significant. Long-term follow-up data were not available from 10 children, which reduced the sample size to two-third ( $n = 21$ ) of the initial sample size. The reduced sample size at

long-term follow-up might not be able to allow a significant effect to manifest clearly. It is also possible that there are factors other than parental relationship quality may predict long-term outcomes. Virues-Ortega & Rodriguez (2013) have found that treatment duration and treatment intensity might be the possible factors that predict the longitudinal growth in behavioural interventions in children with ASD. Nonetheless, the mechanism or the process linking marital relationship and child sleep would require further investigation.

There are several possible explanations for why parental relationship quality would impact on the therapeutic outcomes in children. It is possible that when parental discord exists, the parent-child relationship could also be affected. Meta-analyses have supported the idea that interparental conflict may reduce the quality of parent-child interaction and limit child outcomes (Erel & Burman 1995; Krishnakumar & Buehler 2000). Children with ASD were found demonstrating resistance to sleep interventions when parental discord existed (Hodge et al., 2013). As a result, the therapeutic change of the child might be impaired.

It is also likely that it might be difficult to implement an intervention plan with consistency when the parents have disputes over the treatment process, the child's behaviours or changes in daily routines (Grindle et al., 2009). Inconsistent parenting styles within a family have been found to sustain sleep disturbed behaviours of the children with ASD. Because of that, treatment gains might be reduced (Gau et al., 2010).

Reduced paternal involvement in caring for the children related to marital dissatisfaction might also affect the treatment responses of the children. McBride and Mills (1993) have reported that the level of paternal involvement was relatively high when the father displayed high marital satisfaction or the father's partner also showed high marital satisfaction. Jones and Verduyn (1983) revealed that the attendance of both parents at treatment training sessions could enhance a child's treatment progress in TD children with sleep disturbance. The involvement of both parents has been found associated with positive

outcomes in behavioural problems of children with ASD (Narzisi et al., 2015). Narzisi et al., (2015) also reported that low parental involvement could be the main factor in predicting negative outcomes. Marital satisfaction possibly enhances paternal motivation in supporting the mothers, in pursuing and persisting with the intervention programmes. More support from the fathers might also increase the mothers' marital satisfaction (Johnson & Simpson, 2013) and, as a consequence, the treatment progress of a child is likely to be strengthened.

Another possible explanation is that marital conflicts might alter the biological regulation of a child, such as the hypothalamic-pituitary-adrenal (HPA) activity, which may disrupt a child's sleep. Available evidence suggests that family stressors, such as marital conflict, may negatively affect a child's sleep (El-Sheikh, Erath, Buckhalt, Granger, & Mize, 2008; Sadeh, Raviv, & Gruber, 2000). Literature of child sleep has revealed that a child's exposure to conflict affects the functioning of a child's physiological systems, including vagal suppression to stressors and elevated daytime cortisol level (El-Sheikh et al., 2008; El-Sheikh, Harger, & Whitson, 2001). These might be the indicators of increased HPA activity in response to environmental stressors (Scher, Hall, Zaidman-Zait, & Weinberg, 2010). In TD children, poor sleepers were found to have higher levels of cortisol secretion than good sleepers (El-Sheikh et al., 2008; Hatzinger et al., 2008). Marital conflicts might alter the children's HPA activity and eventually affect the children's sleep quality through a bio-behavioural process. Hence, a home environment with fewer marital conflicts might enhance treatment performance in children.

**Maternal wellbeing and paternal wellbeing.** The results of regression analysis revealed that maternal as well as paternal wellbeing did not predict treatment outcomes in the present study. There are very few outcome studies that treated parental mood states as predictors. The findings of the present study are in contrast to those of existing research which suggests that high levels of parental stress might reduce treatment gains in children

with ASD (Osborne et al., 2008), children with anxiety disorders (Crawford & Manassis, 2001) and children with antisocial behaviour (Kazdin, 1995). One possible explanation for the present findings is that parental mood states and parental sleep were affected by the child's sleep but did not necessarily contribute to the child's sleep problems. A number of previous studies have demonstrated that a child's sleep problems can negatively affect maternal mood and maternal sleep (Chu & Richdale, 2009; Carter, Martinez-Pedraza and Gray, 2009; Doo & Wing, 2006; Meltzer, 2008; Polimeni et al., 2005); however, little is known about the reciprocal nature of this relationship. It is also possible that since the pre-treatment stress, anxiety and depression levels displayed by the parents at pre-treatment were at mild levels, the extent may not suffice to affect the child's sleep substantially.

#### **Treatment Acceptability and Treatment Outcomes Relationship (Research Question 4)**

The fourth research question examined whether treatment outcomes were associated with parental ratings of treatment acceptability. Interestingly, the results of the present study did not show any significant correlation between the TARF-R scores and children's SPS change scores. Treatment acceptability was also found to be unrelated to secondary outcomes. This type of analysis has not previously been undertaken in research into outcome analysis of behavioural sleep treatment in ASD context. Moreover, it is important to examine the factors that contribute to treatment acceptability ratings because parental dissatisfaction with the treatment might increase the chance of dropping out (Kazdin, Holland, & Crowley, 1997). The present study reported that the correlation between acceptability and treatment response is weak. This finding is particularly interesting as it suggests that ratings of treatment acceptability may be dependent upon something other than the therapeutic outcome.

In the present study, parents were provided with free of charge service, flexible scheduling, and a collaborative parents-therapist relationship. There is evidence suggesting

that such processes may increase the ratings of parents in treatment acceptability in research-based treatment (Tharinger et al., 2009). Treatment compliance has been positively correlated with treatment acceptability (Reimers et al., 1992). Those parents who remained in treatment might be more motivated to change than those who dropped out prematurely. In the present study, the parents might assess the treatment acceptability of the intervention based on their willingness to implement the treatment and to change the family habits regardless of the treatment responses of their children.

### **Strengths of the Present Study**

There are several key strengths in the present study worth mentioning. First, currently, the majority of studies have focused on the relationship between child sleep and parental wellbeing (Chu & Richdale, 2009; Hodge et al., 2013; Lam et al., 2003; Meltzer, 2008; Polimeni et al., 2005; Reid et al., 1999; Richdale et al., 2000). Few studies investigated the relationship between family complexity variables and treatment outcomes. The present study contributed toward, and extended previous research in several key ways. This is one of the first studies which has investigated the relationship between family complexity and treatment outcomes. In a review on predictors of outcome in early intervention in ASD, Vivanti et al. (2014) commented that family factors were seldom considered as predictors in outcomes studies. There is no research on examining the possible predictors that reside within the complex family system of a child, and on investigating how well these predictors contribute to the treatment progress of children with ASD. The present exploratory study is the first known study to report the possible parent/family predictors evident in family complexity in ASD and sleep context.

Second, the present study considered the interrelationship among multiple parent variables and treatment outcomes, and as such, moved beyond a univariate approach. This is important because usually only one or two family variables would be included in a sleep

study, which might undermine the joint effects of multiple familial factors on the therapeutic change of a child (Biederman et al., 1993; Kazdin & Whitney, 2006). The identification of three latent components resided underneath multiple family variables suggests that paternal wellbeing, maternal wellbeing and parental relationship quality might be the important domains within the complex family system of a child. In addition, among the ASD sleep studies, the present study was the first to report the finding of mutual satisfaction between two parents as a therapeutic outcome predictor.

Also, the study of Kazdin and Whitley (2006) focused on the effects of case complexity on treatment outcomes in children with an oppositional defiant disorder or conduct disorder. The present study, to the best of our knowledge, is the first study that looked into the possible components that resided underneath the complex family system in children with ASD.

Third, the sample size ( $n = 31$ ) of the present study is substantially larger than the majority of previous sleep studies that have included children with ASD, and thus increasing the chance of important effects or associations to be detected (Tabachnick & Fidell, 2001). As noted by Hogg and Tanis (2009), the statistic values obtained from a sample of more than 30 individuals could be taken as an estimation of the population parameter. Therefore, compared with the findings resulted from studies of small sample sizes, the evidence of the present study provided stronger support for the inference that parent-implemented multi-component behavioural treatments could reduce the sleep problems in children with ASD.

Fourth, one strength of the present study is the inclusion of paternal data whereas most sleep studies have focused on examining the relationship between maternal wellbeing and child sleep (Carter et al.; Chu & Richdale, 2009; Hodge, Hoffman, Sweeney & Riggs, 2013; Hoffman et al., 2008; Lam et al., 2003; Meltzer & Mindell, 2007). Yet within sleep studies in ASD children, rarely have studies examined paternal factors that influence

treatment outcomes (Kazdin, 1995). The inclusion of paternal data in the present study contributes toward our understanding of the fathers' role in child sleep as well as the impact of child sleep on the fathers.

Fifth, long-term follow-up data have been lacking in most ASD studies that used behavioural sleep interventions. This is one of the few studies that collected both short- and long-term follow-up data on treatment responses of children with sleep problems in ASD context (Adkins et al., 2012; Escalona, Field, Singer-Strunck, Cullen & Hartshorn, 2001; Johnson et al., 2013; Knight & Johnson, 2014; Malow et al., 2013; Moore et al., 2004; Reed et al., 2009). The present study provided evidence to the few studies which have collected long-term follow-up data (i.e., of 12 weeks or more post-treatment) in reporting that behavioural treatments could effectively reduce sleep interfering behaviours, and the long-term gains were evident in children with ASD (Durand & Christodulu, 2004; Durand, Gernett-Dott & Mapstone, 1996; Friedman & Luiselli, 2008). Findings of the present study also contributed evidence to the existing research in supporting the long-term efficacy of parent-implemented home-based treatment in reducing sleep problems in children with ASD.

Finally, in the present study, the SPS calculation was performed by two researchers to increase the objectivity. When unclear or suspicious data were found in the sleep diaries, data coding from the relevant video-clips were reviewed by a third research assistant to enhance the accuracy of the data. As a result, the validity of the SPS calculation could be enhanced, and thus the sleep problem severity could be reflected more accurately.

## **Clinical Implications**

Although efforts to choose appropriate intervention strategies are essential for increasing treatment efficacy, it is also important to identify families for whom treatment is

effective. Therefore, it is necessary to understand what familial factors might impair the therapeutic change of a child. From a clinical perspective, identifying any case that is likely to respond or fail to respond would improve the treatment provided to children with sleep problems. The findings of the present study enhance our understanding of what familial factors in the ecological system of a child could influence the treatment efficacy in ASD context; this knowing is beyond the studies in TD children. The identification of ecological niches helps create a desirable environment for individual-environmental interactions to occur (Bronfenbrenner, 1992). The understanding of how sleep correlates with the familial factors helps develop the best fit intervention for alleviating sleep problems in children with ASD.

The present study has extended the scope of family functioning to include marital relationship in the family complexity in ASD context. In a home-based intervention, at least one parent would be a co-therapist (Lovaas, 2003). The present findings suggest that to increase the treatment effectiveness, extra effort might be required to address the parental relationship if marital discord or marital dissatisfaction appear to limit a successful outcome. This provides clinicians with some hints to inform best practice. In clinical practice, the therapist might need to raise the parental awareness of their roles in providing a favourable environment during the intervention process. A home environment with harmonious parental relationship would enhance child-parent interaction, which would eventually enable the full actualisation of a child's potential (Bronfenbrenner, 1992). Existing behavioural treatments might consider a multiple-treatment approach in addressing the child sleep as well as parental relationship to help those children who are less likely to respond.

### **Limitations of the Present Study**

There are a number of limitations that should be considered when interpreting the findings of the present study. Firstly, because more than one-third of the participating



families were single-mother families, paternal data were limited. Consequently, only maternal marital satisfaction was used for the subgroup comparison between high and low marital satisfaction groups. The scarcity of paternal data might limit the generalisability of the present analysis to families with two parents, or to fathers.

Secondly, it should be noted that ten children did not complete the long-term follow-up phase. It is possible that those children who improved less at earlier stage dropped out from the treatment prematurely, and therefore their data are not available for the inclusion in the long-term follow-up analysis. Hence, the long-term effect should be interpreted with caution.

The third limitation is that the information provided by parents in the sleep diaries could be vulnerable to inaccurate or imprecise recording. The sleep diaries were the main source of child sleep data for the calculation of SPS across the four phases. Whilst sleep diaries are a validated method commonly used to collect sleep data, it is possible that some sleep disturbances might go undetected (e.g., extended quiet awakenings) and/or that reporting in a fatigued state could compromise the accuracy of the data. In the larger study, this problem was ameliorated by using video-recording for triangulating the sleep diary data. Nevertheless, the possibility of imprecise manual recording in sleep diary could not be entirely excluded.

The fourth limitation is related to the validity of the SPS composite score. The scoring guide of the SPS composite scores was based on the National Sleep Foundation (NSF) Guidelines (Richman, 1985). However, there is a newly updated version of the National Sleep Foundation Guidelines (Ohayon et al., 2018). The panel has revised the recommended sleep ranges for each child or teenage groups. For instance, the recommended sleep hours range for the teenage group (14 to 17 years old) is widened by one hour (i.e., 8 to 10 hours); it was 8.5 to 9.5 hours in the old version. In the present study, if a teenager had eight hours of

sleep, a score of 1 was assigned. However, if the scoring was based on the new version, a score of 0 would be assigned to the same teenager. Nevertheless, in the present study, the pre- and post- comparison used the same measurement system (i.e., the old version of NSF) for each time point; the difference in scores between any two time points was still a reliable change.

In the present study, the items in the SPS scoring did not include the frequency of curtain calls. Curtain calls are characterised by active resistance, verbal protests, and frequent requests for extra food, drinks or hugs at bedtime (Owens, Chervin & Hoppin, 2017). A high frequency of curtain calls could also reduce the sleep quality of a child (Sciberras, Fulton, Efron, Oberklaid, & Hiscock, 2011). It is possible that the SPS scoring used in the present study has missed out one measure of sleep disturbance which is found common in the families having children with ASD (Cuomo et al., 2017; Kylan & Cynthia, 2012). Therefore, the SPS scoring system could be improved by including the frequency of curtain calls.

The fifth limitation is the sensitivity of the composite family adversity score in detecting the adverse situation of a family. Two of the models in factor analysis indicated that the family adversity score loaded moderately to maternal sleep and maternal mood states. By comparing the models with family adversity variable included in the component of maternal wellbeing and the models without such variable, the findings indicated that models without the family adversity variable accounted for more variance. It is possible that the use of a composite score was not sensitive to case-specific changes related to the family's demonstration of the number of risk factors. The dichotomous scoring method (0 or 1) might have over-simplified the actual adversity condition that the family was undergoing, as suggested by Pheula, Rohde and Schmitz (2011). Nevertheless, in order to minimise the parental burden of filling out a number of assessment forms, and to reduce the risk of pulling

out, the present study did not include other auxiliary measures for assessing each risk factor (e.g. marital conflicts or parental psychopathology) of family adversity separately.

The data used to calculate the family adversity score were mainly based on the summary of clinical interviews and conference discussion with the panel of psychologists involved in the larger study. In the clinical interviews, no standardised questionnaire was used to address the family adversity information or familial risk factors. It is possible that different psychologists might have asked different questions with different focus that might lead to non-systematic answers given by the parents. Therefore, the family adversity data collected might vary across families with different cultures or different perceptions towards the questions. Inconsistency in data collection might limit the reliability of the family adversity score in reflecting the real situation that the family have experienced.

Rutter et al., (1975, 1977) developed a family adversity index that consisted of six risk factor domains (i.e., marital conflicts, low SES, etc.) known to be associated with psychiatric problems and social deviance in young people. The present study is the first attempt in adopting the family adversity framework of Rutter et al., (1975, 1977) in researching the sleep problems in individuals with ASD. It is possible that the nature of sleep disturbance associated with ASD is different from the psychiatric or anti-social nature of TD children, and because of this, the family adversity variable behaved oddly in the present factor analysis. It is also likely that the effect of family adversity might be more obvious if the complexity data were used to predict non-engagement and dropout in the present study. This implies an area for further investigation.

### **Recommendations for Future Research**

In future, it may be interesting to conduct a longitudinal study which investigates the mutual influence between parental functioning and child sleep. This might enable the

possible bi-directional or cyclical relations between child sleep and parental wellbeing to manifest more clearly. The long-term effects of behavioural treatment on child sleep and parental mood states might also be more apparent. Also, it is worth further efforts to include paternal data on child sleep treatment outcomes because paternal influence in child sleep management lacks empirical evidence. The importance of both parents' involvement in the intervention was underlined. Because of this, it would be meaningful for future investigation to examine whether marital satisfaction could enhance parental involvement, and eventually boost treatment outcomes in the context of ASD. Further calls could also be included in the estimation of sleep problem severity to produce a more comprehensive analysis of sleep problems of children with ASD.

The parents in the present study demonstrated obvious reduction in anxiety levels following treatment. However, the available data in the present study were not sufficient to identify the mechanism that enabled the amelioration of parental anxiety during the treatment process. Sleep problems can negatively affect other aspects of a child's daily functioning, such as, learning, behaviour control, sleep and mood regulation (Cohen, Conduit, Lockley, Rajaratnam, & Cornish, 2014; Gozal, 1998; Horne, 1988; Liu, Hubbard, Fabes & Adam, 2006; Schreck, Mulick & Smith, 2004; Quine, 1991). It is, therefore, possible that improvement in child sleep may enhance the daily functioning of the child, and thus the parental worries or anxiety about the child's sleep and wellbeing might decrease (Souders et al., 2009). Future analysis could include daytime behaviours of a child for a better understanding of the possible mechanism underneath parental anxiety reduction.

On the other hand, the parents did not display remarkable attenuation in stress levels in the present study. It is likely that implementing a behavioural intervention of any kind can be a challenging task for some parents (Eikeseth, Klintwall, Hayward & Gale, 2015). Diggle et al. (2002) reported that parents in home-based interventions displayed higher levels of

stress than those in clinic-based treatment in children with ASD. There is some evidence suggesting that parents of children with ASD who implemented in-home interventions displayed increased stress over the course of treatment (Eikeseth et al., 2015). Parents were also found stressed in handling the child's resistance when new strategies were introduced at home (Etherton, Blunden & Hauck, 2016). Therefore, stress level is unlikely to reduce in short-term. The data in the present study were not sufficient to identify factors that might sustain parental stress. Further investigation into any long-term change in parental mood may shed some light on this interesting question.

To minimise the limitations associated with inaccurate or inadequate data recording in sleep diaries, future research could attempt to include objective measures of sleep. Some of the most common objective sleep measurements include actigraphy and polysomnography (Cumon et al., 2017). Goldman et al. (2009) have demonstrated the applicability of using actigraphy to measure sleep (e.g., sleep latency and sleep efficiency) and polysomnography parameters (e.g., total sleep time and arousal index) in defining ASD good sleepers versus poor sleepers. Nevertheless, sleep diaries are sensitive in differentiating whether a child can self-soothe to sleep after a night waking. Therefore, the parent-report sleep diaries and objective sleep measures might complement each other.

With regard to the family adversity score, it might be necessary to develop the final score in a more sophisticated fashion. Among the six indicators identified by Rutter (1975, 1977), some of them can be assessed individually by using relevant psychometrics to inform the subscore of the total family adversity score. For instance, for maternal mental disorder, in addition to a reported history of psychiatric diagnosis, the Self-Report Questionnaire-20 could be used for screening general psychological distress. Similarly, the O'Leary-Porter Scale (OPS, Porter & O'Leary, 1980), which assesses the frequency of marital conflicts that occur in front of the target child, could be used as a proxy for marital discord indicator of

family adversity. This may enhance the validity and sensitivity of the family adversity score in reflecting the disadvantaged factors experienced by a family.

Kazdin and Whitley (2006) proposed that case complexity should contain child complexity and family complexity. Vivanti et al. (2014) have argued that the decision of treatment goals and intervention strategies should be informed by both child characteristics and family characteristics for optimising the treatment outcomes. Personal characteristics (e.g., temperament, gender or age) are linked to the demand and resources of a child (Bronfenbrenner & Morris, 1998). A child brings these characteristics to the social situation in its ecological system. Although the agents in the ecological system can influence a child, the child's characteristics can also actively change the environment. For instance, a child with ASD might demand more time and care from the parents, and the parents might, in turn, invest more time in caring for the child. More parental care and involvement might predict a more positive outcome. The child's functioning, ASD symptom severity, intellectual ability, language and social abilities at pre-intervention were found predictive of treatment outcomes in ASD context (Narzisi et al., Perry et al., 2011; Virues-Ortega & Rodríguez, 2013; Zachor & Itzhak, 2010). For these reasons, future sleep studies might include both child factors and parent/family factors to produce a more comprehensive understanding of the child-family interaction in treatment outcome prediction.

## **Conclusion**

Five key points stand out from the present study. First of all, behavioural intervention demonstrated effectiveness in reducing sleep problem severity in children with ASD. Secondly, secondary outcomes of family-based sleep intervention included the attenuation of maternal anxiety and sleep as well as paternal anxiety and depression. Thirdly, three components of family complexity were found. They were maternal wellbeing, paternal wellbeing, and parental relationship quality. Fourthly, among the three components, parental

relationship quality appeared to be the only significant predictor of children's treatment outcomes. Finally, this is one of the first studies to explore the impact of family complexity on children's treatment outcomes in sleep and ASD literature. While this study has some limitations, the promising treatment effects found in the present study added support to the use of FBA informed home-based multi-component behavioural treatment in helping children with sleep problems in ASD context. For clinical practice, to maximise the treatment outcomes, it is important to consider family relationships when developing a treatment. Future research in these areas is critical in further enhancing our understanding of tailoring best fit family-focused behavioural sleep treatment for children with ASD.

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## **Appendix A: Parent Information Sheets**

### **An investigation into the effectiveness of treatments for sleep disturbance in children with autism or features of autism**

#### **Information for Parents/Caregivers**

This research has been assessed and approved by the University of Canterbury Human Ethics Committee (HEC 2018/47).

Dear Parent/ Caregiver,

We are a group of researchers at the University of Canterbury. Dr Laurie McLay is a Senior Lecturer in the School of Health Sciences at the University of Canterbury. Laurie has many years experience in working with children and young people with developmental disabilities and their families. Associate Professor Karyn France has lectured here for many years, has conducted research into the treatment of paediatric sleep disturbance and is a registered clinical psychologist with considerable clinical experience in this area. Professor Neville Blampied has a similar history of teaching and research. A number of Masters and PhD students and Child and Family Intern psychologists or registered psychologist also work on this project.

We would like you and your child with autism to consider participating in this research study. The primary purpose of this study is to investigate the effectiveness of treatments for sleep disturbance in children with autism. Treatment can include a range of strategies, including both non-traditional approaches (such as white noise) and behavioural interventions. These approaches have been designed to minimise stress as much as possible for the parents and children using them. We are also interested in parents' and children's experiences in using the treatments and any changes to their lives, or their child's lives, which result.

As a part of this study we would also like to investigate the experiences of parents in implementing treatments for sleep disturbance, those treatments that they consider to be most acceptable, and the impact of successful treatment of sleep problems on parent and child wellbeing and quality of life. In order to do this we will ask you to complete some questionnaires about you and your child's well-being and behaviour at the commencement and conclusion of treatment. We will also ask your perspective on the treatment that was provided. We will do this either during visits to your home, Skype interviews, or in a clinic at the University of Canterbury.

If you agree to allow your child to be a part of this study, we will meet with you, or Skype you, to discuss your child's sleep behaviour and find out more about him/her and your family.

This initial meeting will last for approximately 1-1 ½ hours. We will then ask you to complete sleep diaries in which you will record further information about your child's sleep patterns. Sleep diaries will be recorded each day throughout all phases of the study as this will allow us to monitor the effectiveness of the treatment approach. The sleep diaries will take you up to five minutes to complete each night. You will also be asked to complete commonly used questionnaires in order to obtain information about your child's sleep behaviour and the effects of treatment. It will take approximately 15 minutes to complete each questionnaire. When we have established an understanding of your child's sleep behaviour, we will work with you to develop sleep-related goals for your child. This will involve a second treatment planning session which will last 1-1 ½ hours.

To help us gather further information about your child's sleep patterns we will bring or send a video camera to your home for some nights over the course of the programme, which is capable of recording all night sleep. In addition we may ask you, if possible, to use an actigraph with your child. This watch-like device records the movements associated with sleep and can be worn on the wrist or ankle, or secured into a pocket on your child's pyjamas. This may offer an alternative to video cameras when appropriate. These methods will allow us to measure sleep behaviour at times when an adult is not present. We will demonstrate and explain how to use each of these methods for gathering information.

When information about your child's sleep behaviour has been gathered, treatment will commence. You will be offered a choice of treatment options, which you will then implement with the support of the research team. If you are dissatisfied with the treatment approach or the degree of progress that is being made then you will be offered a choice of another treatment option. We will provide you with all of the necessary information about each treatment approach and we will maintain regular contact with you during treatment. It is anticipated that your involvement in the study will occur over the course of a few months, but will depend on the rate of your child's progress as well as your satisfaction with the progress.

For the purpose of this project, myself (insert name), a psychologist/intern psychologist/Masters/PhD student will be working closely with you to conduct the necessary assessments and formulate interventions. XX, a research assistant/Masters/PhD student who also works as a part of our sleep team, may look at some of the information that we collect, such as video recordings and actigraph data.

Your child will be assigned a code name to ensure anonymity and anything that you or your child says or does will be kept confidential. The results of the study may be submitted for publication to national or international journals and may also be presented at conferences. No identification of the child or family will be possible from any report, publication or presentation.

If you want to withdraw from the project before completion, you can do this at any time without penalty or repercussions.

Should you require any additional information about the study or if you would like to access the study findings you are able to do so at any stage. The data which is produced from the research will be kept in a locked cabinet at the University of Canterbury for a minimum of ten years.

If you agree for your child to take part in the research, please sign the consent form that is attached.

If you have any complaints you may contact the Chair of the University of Canterbury Ethics Committee. The contact details are given below.

If you have any questions about this project please feel free to contact Dr Laurie McLay: Phone (03) 369-3522 or, email: [laurie.mclay@canterbury.ac.nz](mailto:laurie.mclay@canterbury.ac.nz)

## **Appendix B: Parent Consent Form**

### **An investigation into the effectiveness of treatments for sleep disturbance in children with autism**

#### **CONSENT FORM FOR PARENTS/ CAREGIVERS**

This research has been assessed and approved by the University of Canterbury, Human Ethics Committee (HEC 2018/47).

- ☐ I wish to participate in the project, “An investigation into the efficacy of treatments for sleep disturbance in children with autism”
- ☐ I have read and been given a full explanation of this project and have had the opportunity to ask questions.
- ☐ I understand what will be required of myself and my child/the child in my care during this project.
- ☐ I understand that the investigators do not foresee any potential risks to me or my child as a result of participating in this study. However, if the intervention results in an increase in family stress, the staff working with us will provide support.
- ☐ I understand that all information about my family will be treated as confidential unless there is concern about anyone’s safety. In this case my clinician will need to speak to someone else to ensure the safety risk is removed. No findings that could identify me or my child will be published
- ☐ I understand that the findings of this study may be published in a research journal or at a conference and that the anonymity of my child and I will be maintained
- ☐ I understand that participation in this project is voluntary and that I can withdraw my child or he/she can withdraw from the project at any time without repercussions. I can also withdraw any data that has been collected at any time prior to the publication of that data



- ☐ I understand that all research data that is collected will be securely stored at the University of Canterbury for a minimum of ten years
- ☐ I understand that I am able to request a copy of the results of this research, should I wish to do so, and that these results will be provided for me
- ☐ I allow video-taping of my child's sleep behaviour to be completed by the researcher and understand that this videotape will be used for data gathering purposes only. I also understand that I have the right to request that video footage is destroyed at any stage.
- ☐ I consent to others, listed below, being involved in the implementation of the intervention

Name: \_\_\_\_\_

Date: \_\_\_\_\_

Signature: \_\_\_\_\_

Others I consent to implementing intervention:

Name: \_\_\_\_\_

Name: \_\_\_\_\_

Name: \_\_\_\_\_

- ☐ I would like a summary of the results of this project.

***Please return this form to XXX.***

## **Appendix C: Child Information Sheet**

### **An investigation into the efficacy of treatments for sleep disturbance in children with autism**

#### **Child Information Sheet**

Hello. My name is XX and I am a XX student at the University of Canterbury. I am doing a project about how to help children to sleep better and I would like for you to help me with this.

I am going to be talking to you and your parent/s about ways to help you to sleep better. This means that I might be Skyping you, coming to your house, or your parent/s will be coming to see me at the University.

There will be a video camera in your bedroom sometimes. This will help me to understand what you do when you are awake and asleep. Only your parents and other people working on this project will be able to see this video. We may ask you to wear an actigraph. An actigraph is worn on your wrist like a watch and it tells us when you are asleep and when you are awake.

If you do not want to be a part of this project, you can tell me or your parents and you won't need to be a part of it anymore.

If you have any questions you can ask me or your parents whenever you like.

Now we need to decide if you would like to do this. If you do want to be a part of my project then you can say "yes". If you do not want to be a part of this project then you can say "no" and no one will mind.

If you say yes, you or one of your parents can sign the form for you.

**Appendix D: Child Consent Form**

**“An investigation into the efficacy of treatments for sleep disturbance in  
children with autism”**

**Child Consent Form**

My name is \_\_\_\_\_.

- ☐ XXX has told me about the work that she is going to be doing with me and my parent/s.
- ☐ XXX told me that she is going to be working with me and my parent/s to help me to learn to sleep better.
- ☐ While XXX does this she will be asking my parents about my sleep each night and there will be a video camera in my room on some nights that is recording my sleep.
- ☐ I know that if at any time I want to stop being a part of this project then XXX will stop recording data and this will be destroyed.
- ☐ If I want XX to stop video recording my sleep then the camera will be taken out of my room and that will be fine. If I want any video footage to be deleted, I can tell XXX or my parents.
- ☐ I was told that my parents/caregiver may sign this form for me and I think that is OK.
- ☐ I would like a summary of the results of this project.

Child's name: \_\_\_\_\_

Date: \_\_\_\_\_

Signature: \_\_\_\_\_

If this form is signed on behalf of your child please acknowledge, by signing this form, that your child was verbally informed of the investigation and what it will involve and that they were unable to provide verbal or written consent that they would like to be a part of this research.

Parent/caregiver: \_\_\_\_\_

Date: \_\_\_\_\_

Signature: \_\_\_\_\_

***Please return this form to XXX.***

## Appendix E: Child Assent Form



The project that XXX wants to do to help me with my sleep has been explained to me. I know I don't have to be a part of it if I don't want to. If I have any questions I can ask XXS or my parents.

- I am happy to be a part of the project and for XXX to help me with my sleep so I have coloured in the happy face.

OR

- I don't want to be part of the project or to have any help with my sleep so I have coloured in the sad face.



My name: \_\_\_\_\_

You can give this form back to XX now.

## **Appendix F: Example of Clinical Interview Questions**

*Begin with confidentiality terms and obtain consent to be interviewed;*

### *Problem behaviour*

- Tell me about what your concerns are?
- Walk me through a typical night?
- Problem behaviour; frequency, duration, setting, when does it occur, when does it not occur, what happens immediately after the behaviour, what makes it better/worse, what do you do when the behaviour occurs?
- Opinion- what do you think causes the behaviour?
- Sleep hygiene- what is the current bed time routine, time and place of bed, wake up time

### *History of problem behaviour*

- Approximately how long has the behaviour been occurring?
- Have there been changes in the frequency or intensity of behaviour over time?
- What attempts have been made in the past to change the behaviour? Were they successful?

### *Developmental History*

- How was the pregnancy, birth, parent reactions to having a child at the time?
- What was his behavioural style like as an infant?
- Did he meet all his milestones?
- Who are the significant people in his life?
- Do you have any concerns for him, aside from sleep?

### *Families of origin*

- How do you think your own backgrounds have impacted on your parenting style?

- What supports do you have in place at the moment?
- Other questions related to the parental wellbeing and significant incidence happened in the family.

*Summary and termination*

## **Appendix G: SPS Scoring Guide for School-aged Children**

### **Sleep Problem Severity Score Scale – Preschool-aged (3 - 4yrs 11m)**

\* Based on Richman, (1985) and National Sleep Foundation Guidelines

#### *Time taken to fall asleep each night*

<15	0
16-29	1
30-44	2
45-60	3
> 60	4

#### *Total time slept each night in hours*

10+	0
9+	1
8+	2
7+	3
6+	4

#### *Bedtime each night (whichever is worse)*

5 – 8.4pm	0
8.5 – 9.2pm	1
9.3 – 10pm	2
10.1 – 11pm	3
After 11pm	4

#### *No. of wakings each night*

0	0
1.0	1
2.0	2



3.0	3
4+	4

*Average time awake per waking (min) each night*

0 – 5	0
6 – 15	1
16 – 30	2
31 – 60	3
>60	4

*Percentage of night spent co-sleeping each night*

None	0
1 – 20	1
21 – 40	2
41 – 60	3
61+	4

*Parental presence for initial sleep onset*

Not present	0
Present	1

*Parental presence during sleep onset following night waking*

Not present	0
Present	1

## Appendix H : SPS Scoring Guide for School-aged Children

### Sleep Problem Severity Score Scale – School-aged (5 – 12 years)

\* Based on Richman, (1985) and National Sleep Foundation Guidelines

#### *Time taken to fall asleep each night*

<15	0
16-29	1
30-44	2
45-60	3
> 60	4

#### *Total time slept each night in hours*

9+	0
8+	1
7+	2
6+	3
5+	4

#### *Bedtime each night (whichever is worse)*

Up to 9.2pm	0
9.3 – 10pm	1
10.1 – 11pm	2
11.1 – 12pm	3
After 12pm	4

#### *No. of wakings each night*

None	0
1	1
2	2
3	3

4+	4
----	---

*Average time awake per waking (min) each night*

0 – 5	0
-------	---

6 – 15	1
--------	---

16 – 30	2
---------	---

31 – 60	3
---------	---

>60	4
-----	---

*Percentage of night spent co-sleeping each night*

None	0
------	---

1 – 20	1
--------	---

21 – 40	2
---------	---

41 – 60	3
---------	---

61+	4
-----	---

*Parental presence for initial sleep onset*

Not present	0
-------------	---

Present	1
---------	---

*Parental presence during sleep onset following night waking*

Not present	0
-------------	---

Present	1
---------	---

## Appendix I : SPS Scoring Guide for Adolescents

### Sleep Problem Severity Score Scale – Teens (13 years+)

\* Based on Richman, (1985) and National Sleep Foundation Guidelines

#### *Time taken to fall asleep each night*

<15 mins	0
16-29	1
30-44	2
45-60	3
> 60	4

#### *Total time slept each night in hours*

9+	0
8+	1
7+	2
6+	3
5+	4

#### *Bedtime each night (whichever is worse)*

Up to 9.2	0
9.3 – 10	1
10.1 – 11	2
11.1 – 12	3
After 12pm	4

*No. of wakings each night*

None	0
1	1
2	2
3	3
4+	4

*Average time awake per waking (min) each night*

0 – 5	0
6 – 15	1
16 – 30	2
31 – 60	3
>60	4

*Percentage of night spent co-sleeping each night*

None	0
1 – 20	1
21 – 40	2
41 – 60	3
61+	4

*Parental presence for initial sleep onset*

Not present	0
Present	1

*Parental presence during sleep onset following night waking*

Not present	0
Present	1

### Appendix J: Sleep Diary Template

	Date:	Monday:	Tuesday:	Wednesday:	Thursday:	Friday:	Saturday:	Sunday:
Daytime sleep	Setting (where fell asleep)							
	Time asleep							
	Time awake							
Night-time sleep	Setting (where fell asleep)							
	Time put to bed							
	Frequency of Curtain calls*							
	Curtain calls after put to bed (Describe each)							
	Your responses to each curtain call (Describe each)							
	Best estimate of time asleep							

Child's Name:

[illegible]





**Appendix K: Video Recording Consent Form**

**An Investigation into the Efficacy of Treatments for Sleep Disturbance in  
Children with Autism**

**AUDIOVISUAL RECORDING CONSENT FORM**

You have been given this form because the researchers have asked your permission to take audiovisual recordings of your child's sleep behavior.

Please read the statements below, which explain the purpose of audiovisual recording and how your privacy will be protected:

- The purpose of recording is to gather data for the research project
- Audiovisual recording will only be done with your knowledge and consent
- You can withdraw your consent to audiovisual recording at any time, without having to provide a reason for changing your mind
- The audiovisual file will only be seen by the researchers
- The audiovisual recording will be securely stored at the University of Canterbury for a minimum of ten years

I hereby consent to audiovisual recordings being made on the above conditions.

Signed: \_\_\_\_\_

Date: \_\_\_\_\_